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THE SENSITIVITY OF INFANTS WITH SPINA BIFIDA TO SENSORY INFORMATION

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Doctor of Physical Therapy Program
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ABSTRACT

The Sensitivity of Infants with SB to Sensory Information
Katie Gulsvig, Christina Hawn, James Plummer, Ann Schmitz

Advisor: David Chapman, PT, PhD

BACKGROUND AND PURPOSE: Spina bifida (SB) is the most common neural tube defect in the United States. These babies move their legs less often and demonstrate motor milestones significantly later in life than their typically developing (TD) peers. Research has shown TD infants move their legs more or less often depending on the quality and quantity of sensory information they have available to them. Significant milestones such as crawling and walking rely on intra-limb coupling, which occurs when one limb adapts to contextual environmental factors depending on the position of the other limb. Current motor development processes are rooted in theoretical concepts from the Dynamic Systems Theory and the Theory of Neuronal Group Selection. The purpose of this study was to determine if infants with lumbar or sacral SB would move their legs more or less often when confronted with changes in sensory information while seated in an infant seat designed to facilitate movement.

METHODS: Infants with SB were videotaped in an infant seat designed to facilitate leg movements when they had 0%, 25%, 50%, 75%, and 100% of their estimated calf mass attached to one leg. Leg order and weightings were randomized. Frame by frame behavior coding was used to determine the frequency of leg movements for each condition. Data were analyzed to determine sensitivity to the weighted conditions over developmental time.

RESULTS: Infants in our study demonstrated significantly more leg movements in 5 out of 8 of the weighted conditions compared to baseline. The infants showed a significant change in frequency of leg movements, percentage of left leg movements, and percentage of right leg movements over developmental time. There were also significant negative correlations between thigh and calf skin fold values as well as plantarflexion range of motion and the frequency of leg movements.

CONCLUSION: These results suggest that infants with SB may be less sensitive to sensory information or may be utilizing this information differently compared to TD infants. Simply adding small weights to the limbs of infants with SB can increase the frequency in which they move their legs.

The undersigned certify that they have read, and recommended approval of the research project entitled:

THE SENSITIVITY OF INFANTS WITH SPINA BIFIDA TO SENSORY INFORMATION

submitted by
Katie Gulsvig
Christina Hawn
James Plummer
Ann Schmitz

In partial fulfillment of the requirements for the Doctor of Physical Therapy Program

Primary Advisor: *David Chapman*   Date: 4/26/12

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Introduction and Review of Literature

The physical therapy literature is limited regarding the developmental profiles displayed by infants with spina bifida (SB). Thus, it is often difficult for physical therapists and other healthcare providers to fully comprehend the developmental implications of the primary pathophysiology and associated secondary impairments experienced by these infants. It is also challenging to find evidence-based treatment options for these infants and children. As a result, our goal for this pilot study was to expand the literature regarding how infants with lumbar or sacral SB process sensorimotor information as they learn to move their legs over developmental time. Our aim was to examine how infants with lumbar or sacral SB adapt to changes in their sensory information when small amounts of weight were applied to their legs. We begin with an overview of SB and related pathophysiology.

Spina Bifida is the most common severely disabling neural tube defect (NTD) in the United States. Malformations in the spinal cord and posterior vertebral arches typically characterize SB. Spina bifida occurs during the fourth week of embryogenesis and affects about 1 out of every 2000 live births.[1] Most cases of SB are diagnosed prenatally via blood testing of the mother, ultrasound, and/or amniocentesis. However, some cases go undiagnosed until birth. In these situations, SB can be detected via x-ray, MRI, and/or CT scan.[2]

Researchers and doctors do not know the exact reason(s) why this occurs. Several risk factors, such as, race (SB is more common among whites and hispanics), family history of NDT's, folate deficiency, selected medications, if

taken during pregnancy, eg, anti-seizure medicines, uncontrolled diabetes, obesity, increased body temperature early on in pregnancy have been identified (http://www.mayoclinic.com/health/spina-bifida/DS00417/DSECTION=risk-factors). It is important to note that if a woman takes 400 mcg of folic acid one month prior to conception as well as during the first trimester the chance of having an infant with SB or any other NTD is reduced by 70%.[3] The exact role folic acid plays in preventing SB is also unknown.

There are three types of SB, including occulta, meningocele, and myelomeningocele (MM).[4] Spina bifida occulta is the mildest form of spina bifida, occuring in 10-20% of otherwise healthy individuals.[3] It is characterized by incomplete closure of the vertebral column, however the gap in the vertebral column is too small to allow for spinal cord protrusion. The skin at the site of this pathology may be normal, display a hairy patch, dimple, or birthmark. Many people with this form of SB are unaware they have it as they are often asymptomatic.[3]

Meningocele is the least common form of SB. This type of SB is also characterized by a gap in the posterior vertebral arches. In this case, the gap is wide enough for the meninges to push through without nervous system tissue being exposed. Lesions of this type may or may not be covered by a layer of skin. Symptoms may vary from none to those associated with more complex NTD's (http://spinabifidainfo.com/meningocele-vs-myelomeningocele/).

Myelomeningocele is the most common and most serious type of SB. It results from incomplete closure of the posterior vertebral arches in the spinal

column. This opening allows the spinal cord and meninges to protrude posteriorly out of the spinal column onto the surface of the child's back. This usually results in moderate to severe levels of impairment of the motor and sensory nerves along with the structures they innervate (http://www.ncbi.nlm.nih.gov/pubmedhealth/PMH0002525/). Lesions of this type can occur anywhere along the spinal column, but are most common in the lumbar and sacral regions of the spine with approximately 75% of all MM's occurring in these region of the spine.[5] Note, if there is a MM it is usually repaired within the first 24-48 hours after birth.

The level of the lesion can significantly impact the amount of sensorimotor disruption experienced by infants with SB. Higher level lesions, of course, lead to greater levels of sensorimotor impairment and may impact multiple areas or domains of the child's life. For example, infants with SB may show signs of impaired motor function, bowel and bladder sphincter control, hydrocephalus, and infections of the central nervous system (CNS).[6]

The level of the lesion can also be used to "predict" motor outcomes.[5] For example, about 90-100% of individuals who have sacral lesions become community ambulators and have fewer co-morbidities than their counterparts with higher level lesions.[5]

There are multiple secondary impairments that are often experienced by infants and children with SB. These may or may not require additional surgeries to repair. For instance, infants and children with SB who experience

hydrocephalus usually require surgical placement of a ventricular-peritoneal (VP) shunt. A list of common secondary impairments is presented in Table 1.

Table 1.

Secondary Impairments Associated with SB

Hydrocephalus
Musculoskeletal Impairments: equinovarus, complications with disuse
Bowel and bladder issues
Tethered Cord
Seizures
Vision Problems
Skin Issues; eg, latex allergy
Learning disabilities: difficulty paying attention, problems with language and reading comprehension, and trouble learning math

Many of these secondary impairments, in conjunction with the spinal lesion, commonly result in altered levels and/or quality of sensory information infants with SB have available to them as they learn to generate coordinated movements, like reaching, kicking, and pulling to a stand.[7] In addition, these types of impairments may make it difficult for them to integrate the sensory information that they have available to them. To date, no one has examined in controlled observations the sensitivity of infants with SB to sensory information nor has anyone shown that infants with SB demonstrate different levels of sensitivity to sensory information in each of their legs.

Chapman was among the first to report that infants with SB appear to have significant disruptions in their sensorimotor pathways.[7] Sensorimotor processes are:

"integral to the awareness of stimuli, integration of stimuli into meaningful perceptions of environmental events, and access to physical and social environments that enable infants to actively explore, and engage in meaningful activities" (p. 646).[8]

Therefore, infants and children with SB may experience altered perceptions of their sensory information that might interfere with their ability to achieve important motor milestones, such as crawling and walking.[8]

To achieve coordinated, alternating leg movements that characterize the crawling and walking, an infant needs to be able to rely on some level or type of adaptive or adjustable intra-limb coupling. For example, Ulrich et al reported that when they added small amounts of weight to the legs of typically developing (TD) infants and infants with Down syndrome (Ds) they found they both groups of infants maintained their baseline frequency of leg movements by moving the weighted leg less often while they moved the un-weighted leg more often when seated.[9] What is interesting about these results is that both groups of infants had to overcome their intrinsic tendency or natural preference to move one leg when they moved their other leg. That is, in the baseline condition with no leg weights added these infants usually moved their right leg when they moved their left leg and vice-versa. These types of intrinsic tendencies or natural preferences have been described as intrinsic dynamics.[10]

Intrinsic dynamics refer to the collective behavior of the infant – in our case spontaneous leg movements in the absence of any specific task requirements or task matching.[7,10] Inter-limb coupling of the legs is one example of intrinsic dynamics. In this case, one leg tends to move at a frequency that is

similar to the other leg. As noted above, Ulrich et al demonstrated this concept when they reported that infants with and without Ds altered the frequency of the weighted and un-weighted leg to maintain their overall frequency of their leg movements compared to the baseline trials during which no leg weights were added to either leg.[9]

For this to occur, these infants needed to overcome their intrinsic dynamics or the preference they exhibited during the baseline trials in which they tended to move one leg when they moved their other leg. These observations suggest that these two infant groups were able to adapt to changes in their movement context, i.e. when small weights were added to one leg by altering how often they moved the weighted and un-weighted leg, respectively. Through long term follow up these researchers were able to show that infants who were more or less sensitive to the leg weights walked earlier and later in life, respectively. These results highlight that all infants possess unique intrinsic dynamics as well as how changes in the movement context may influence the acquisition of functional motor skills, like pulling to a stand and walking. These data also suggest that infants with SB who appear to lack sensitivity to sensory information may have altered intrinsic dynamics that may explain, in part, the delays they experience in gaining new motor skills compared to TD infants. The developmental milestones for TD and infants/children with SB are presented in Table 2.

Table 2.

Developmental Motor Milestones of TD Infants and Infants with SB

	TD infants	Infants with SB
Head Control	3 months	Not Established
Rolling	4 months	Not Established
Sitting	6-7 months	1-2 years
Crawling	7-11 months	1-2 years
Standing	9-13 months	3 years
Walking	12-14 months	3-7 years

Motor Development

The developmental profiles of infants and children have been formally studied for nearly 60 years. Multiple theories, including neuro-maturation, ecological psychology, and behaviorism have been employed by a variety of researchers over time as they worked to first describe and then explain how infants and children gain new motor skills.[11-14] Most recently, however, principles from dynamic systems theory (DST) and the Theory of Neuronal Group Selection (TNGS) have been used by a variety of researchers to examine the developmental sequence that infants and children with and without disabilities go through as they seek to acquire new motor skills.[15-20]

Proponents of this approach to studying motor development believe that infants and children *self-organize* their multiple sub-systems, e.g. muscles, peripheral nerves, the central nervous system (CNS), motivation, the environment and the child's movement history, as they learn to generate coordinated movements.[19-23] Thus, new motor skills are thought to emerge over time as a result of the dynamic and cooperative interaction of the infant or child's multiple sub-systems. As a result, the motor behaviors observed at a given point

in development, e.g. today or tomorrow, reflect this on-going dynamic interaction within a given movement context. In addition, systems' theorists suggest that no one system is more or less important to the performance of a particular motor skill. Rather, all systems are thought to contribute to movements that meet the demands of a specific task. Rolling over, for instance, requires a certain amount of motivation, nervous system input to the appropriate muscles, enough muscle strength in the arms, legs, head, and trunk to overcome inertia and gravity, and a relatively flat surface, etc. The lack of sufficient development in one or more of these systems may act as a *rate limiter* and prevent the infant from successfully rolling over.

Self-organization means that highly ordered patterns of behavior, such as alternating leg movements, can emerge from the cooperative interactions of multiple systems in a given context without explicit instructions to meet the demands of a task.[18-20,23] This principle places equal importance on the organism and the environment. As a result, neither has priority in explaining the observed behavior(s). There are several examples that illustrate this principle. For instance, Thelen showed that 6-7 month old babies, who demonstrated no stepping capabilities when held on a firm non-moving surface were able to generate alternating steps when they were supported over a small motorized treadmill (TM).[24] This example illustrates that infants even though they may not consciously understand the task or purpose of stepping are able to organize their multiple sub-systems to meet the demands of the task. These observations support the idea that motor behaviors are not intrinsically hard-wired. Rather,

they result of the cooperative interactions of the infants' multiple sub-systems in a given environment and highlight the important and influential role that the context plays in motor development.

One of the more remarkable examples of self-organization was provided by Thelen et al, in 1984.[25] This group of researchers held non-ambulatory 7-month old infants with limited spontaneous stepping behaviors over a split-belt TM. These infants were produced alternating steps when the TM belt was turned on. More importantly, they were also able to generate alternating steps when one of the TM belts was moving at twice the speed of the other TM belt. The infants accomplished this by adjusting their stance and swing phases to meet the demand created when one TM belt was moving faster than the other TM belt. In addition, this group of infants were able to increase their step rate by 50% when the treadmill belt speed was doubled. These results also reveal the ability of young infants to self-organize their multiple sub-systems to meet the demands of the task and show that early in life infants have some type of inter-limb coupling between their left and right legs.

A final example of self-organization is provided by Chapman when he demonstrated that leg movement frequency can be influenced by changing the movement context for infants with and without SB.[7,23,26] He examined the frequency of leg movements and kicks generated by six TD infants and six infants with lumbar or sacral SB over four months developmental time. The three movement positions were supine, seated in a conventional infant seat, and seated in a specially designed infant seat. Chapman reported that infants in both

groups moved their legs more often and generated more kicks when they were seated in the specially designed infant seat compared to when they were supine or seated in a conventional infant seat. They also altered the velocity and overall range of motion of their leg movements when they were placed in each of the three movement contexts. This study supports the idea that young infants are capable of adjusting the quantity and relative quality of their movements depending on the movement context in which they are placed. Further, these results highlight the importance of multiple systems contributing to the movements we observe.

Each of these papers support the idea of self-organization. In each example, the involved babies adjusted their movements as they were placed in different movement contexts. To be able to do that, the infants had to rely on plasticity within their nervous systems. If they did not possess some type of neural plasticity they would not have been able to adjust their movement patterns. Rather, they would have moved in the same way regardless of the environment or demands of the task that they had to confront. In the TM papers, the infants would not have altered their rate of stepping or been able to adjust to the two TM belts moving at different speeds nor would they have been capable of moving their legs at different rates and speeds when placed in different positions.

The second principle of DST is the concept of a control parameter. Simply stated, a control parameter is a component of the system that acts as a primary agent of change, that is, one that moves the system through a phase shift to a new behavior. [7] Thelen's 'fish tank' experiment illustrates this principle.[27] She and

her colleagues examined the stepping response of a small group of newborn infants by holding them on a firm surface. Then, she asked the parent(s) to bring their infant back to the laboratory when they no longer spontaneously generated steps when held over a table or firm surface at home. During the second visit to the lab, Thelen held the babies on a firm surface to demonstrate that they no longer showed any signs of the newborn stepping response. Next, she held the babies in a fish tank of warm water so that the infants were submerged to about chest height. During this phase of the experiment she video-taped the leg movements of each infant. She found that all of the babies produced alternating steps when held in the fish tank of water. Thelen also reported that infants who had gained the most weight and had the largest thigh and calf skinfold generated the fewest steps and showed less hip and flexion when they did step while held in the fish tank. These results illustrate the important role the environment can play, but also show how select factors, in this case, the biomechanically properties of the infants' legs influence the behaviors they produce and we observe.

A second example of a control parameter is provided by the papers referenced earlier by Chapman.[7,23] He showed that the context can function as a control parameter when he reported that infants with and without SB moved their legs more often and altered the quality of their leg movements when they were seated in a specially designed infant seat compared to when they were supine or seated in a conventional infant seat. Thus, for these infants their position in

space served as a control parameter for increasing the frequency of leg movements and generating more frequent kicks over developmental time.

In contrast to control parameters are rate limiters. A rate limiter is an element or subsystem that is prevents the mover from moving in a certain manner.[7,23] From the examples mentioned earlier, we have shown that rate limiters may be biomechanical factors, i.e. changes in posture, increases in the amount of adipose tissue on the legs; neural factors, e.g. nervous system maturation; and/or not modifying the movement environment to accommodate for changes within the infants' bio-physical sub-systems. All of these examples show how a given element or sub-system may function to limit the motor behaviors that infants and children are able to demonstrate, especially infants who have some type of neurological impairment or disability, e.g. infants with Ds or SB. They also show how we can, by manipulating one or more environmental factors, influence the motor behaviors demonstrated by infants with a wide range of abilities.

Collectively, these studies do not help us understand the relationship between the CNS and other systems, such as the musculoskeletal system, of the human body. In fact, principles of DST do not account for, nor do they explain how the CNS develops. Thus, we turn our attention to the Theory Neuronal Group Selection as put forth by Edelman.[15-18]

Edleman posits that the central nervous system (CNS) interacts in a reciprocal fashion in real time with other subsystems of the body, musculoskeletal, integumentary, cardiopulmonary systems, etc.[15-17] Edelman suggests that the nervous system, specifically groups of neurons, are continually

strengthened throughout an infants' life as they move to meet the movement demands they encounter in various settings. For example, when an infant is learning to kick their legs, the neural connections that support kicking are strengthened as the infant repeats that behavior over developmental time. Likewise, as a baby or child learns to produce coordinated reaching movements the infant will, over time, strengthen the neural connections that supports reaching. Repeated behaviors then strengthen neural connections that lead to the formation of neural maps that support the infants' ability to produce a given type of behavior, e.g. kicks or reaches, in a variety of settings over developmental time. This on-going interactive process influences or shapes the infant or child's movement repertoire as the infant grows and explores the world.

Alternatively, movements that are not repeated often or rarely used will not lead to strengthened neural connections that support those behaviors. Thus, movements that are not reproduced frequently will result in neural connections that become relatively weakened over time. This too, will exert an influence on the breadth and depth of movements a given child demonstrates.

Clearly, because no two individuals experience life in the same manner the behaviors generated and the neural connections that support those behaviors are unique to each person. In addition, because of this neuro-developmental process, movement behaviors can be shaped through different environmental and movement experiences throughout life.[7] For example, infants with and without SB who are placed in a typical infant seat, would not according to TNGS, strengthen the neural connections needed to support efficient and coordinated

hip/knee flexion and extension. This is because this type of seat inhibits those types of movements which would lead to weakened neural connections for hip and knee flexion and extension over time.[7,23] However, if infants are placed in environmental contexts, e.g. held on a small motorized treadmill or seated in a specially designed infant seat, that encourages hip and knee flexion and extension they will be afforded the opportunity to strengthen the neural connections that support those movements over time. This would increase the probability of them producing these types of purposeful, efficient hip and knee flexion and extension movements in the future.[7] In fact, Ulrich and Ulrich demonstrated this concept when they shared that the frequency of leg movements was directly related to when in life infants with Ds learned to walk.[28] They reported that early walkers were likely to kick more often during infancy compared to toddlers with Ds who walked later in life.

As stated above when goal direct movements are repeated over time the neural circuits that support those movements become strengthened. Over time and with varied movement experiences infants learn to modulate their intrinsic dynamics to produce coordinated and efficient movements that meet the demands of the task and context. This allows them to develop their own unique neural map. This type of neural development may serve as a control parameter or rate limiter in the development of specific motor skills.[4] Thus, parents and therapists need to remain sensitive to the impact different movement contexts have on their child's ability to move.

Taken together, principles from DST coupled with TNGS provide us with a logical rationale to examine how infants and children with SB learn to produce coordinated movement. It also offers us a way to explain the unique combination of characteristics that each infant "comes with" and in turn, how we as healthcare providers can examine and intervene to facilitate motor skill development of each infant and child with SB.

Although infants with SB usually present with compromised nervous systems, they too experience the benefits of the reciprocal relationship that exists between their nervous system and their other sub-systems. This idea combined with the principles of self-organization and control parameters makes it probable that we can first test and then develop effective interventions for infants and children with SB. Efforts like this can be designed to help them learn to generate movements that meet the demands of the task in particular environments and that will facilitate their ability to produce functional goal directed movements over time. Of particular interest to us, as physical therapists, is how infants develop the ability walk. Thus, we are motivated to examine how we can influence the leg movements infants demonstrate prior to walking.

Ambulation is a major motor milestone for most parents and children. Previous research has shown that the frequency of leg movements is directly correlated with when in development infants and children learn to walk.[28] Leg movements in TD infants are seen as a precursor to ambulating. These movements have been observed as early as 16 weeks gestational age. Infants with SB also demonstrate in-utero leg movements.[29,30]

Research studies have shown that 16-24 week old fetuses with lumbar or sacral SB detected via ultrasonography, displayed similar frequency and quality of leg movements as TD infants in-utero. Ultrasound studies to 16-24 week old infant with thoracic, lumbar, or sacral spinal lesions have demonstrated the ability to flex and extend their hips as frequently as TD infants.[29,31] Similarly, Sival et al reported that fetuses 18-39 weeks old with thoracic or lumbar lesions, observed via ultrasound, generate leg movements which appeared to be of normal amplitude and speed compared to TD fetuses.[29] However, these leg movements diminish shortly after birth. Studies by Sival have demonstrated that this occurs in as soon as one to seven days following birth.[29] These leg movements in addition to occuring fewer in frequency, also showed reduced amplitude, variability and direction.[32,33]

Rademacher, Black, and Ulrich analyzed leg movements of infants in supine with lumbar or sacral SB at one, three, and six months of age.[34] Results of this study demonstrated that these infants when placed in the supine position, showed depressed movement activity, noted through producing fewer frequency of leg movements when compared to their TD age-matched peers. The results of this study also showed that the younger infants (in this study one and three months) produced the fewest number of leg movements compared to the six month old infants. These findings are supported in other research studies comparing the frequency of leg movements in infants with SB.[34]

Chapman's research has shown that infants with SB tend to move their legs less often than their TD peers.[7,23] This may be due to diminished sensory

information that they have available as a result of their spinal lesion. If so, it is likely that infants with SB move and kicks their legs less often than TD infants because it is a relatively 'un-rewarding' activity, that is, that do not experience a high level of quality or quantity of sensory information relative to their TD peers which results in fewer leg movements and kicks. These observations coupled with the concept of neural plasticity and the report that how often infants move their legs and kick is directly linked to when they learn to walk suggests that we need to develop strategies that will enhance the level of sensory information infants with SB have available to them as well as interventions designed to help them move their legs more often.[7,23,26,35]

Knowledge about the positions or contexts that help babies move their legs more frequently will offer parents and healthcare professionals an opportunity to facilitate earlier ambulation. The current concepts supporting motor milestone development in infants and children are rooted in DST, which states that a change in the environment or movement conditions, may cause a change in the observed behavior(s). This offers a direction for studying these kinds of behaviors. However, it is unknown how infants with SB will respond to manipulations of their sensory information. Will babies with lumbosacral SB move their legs more or less often when small amounts of weight are placed on them? Will they alter how often they move the weighted leg versus their un-weighted leg like TD and infants with Ds do?[9] Is there an optimal amount of weight that can be added to the infants' legs that will result in more leg movements than other levels of weight? Thus, the purpose of this study is to determine if infants with lumbar

or sacral SB, between four and 12 months of age at entry into the study, move

their legs more or less often when they have 25%, 50%, 75%, and 100% of their

calf mass added to one of their legs while they are seated in an infant seat

designed to facilitate leg movements over three months of developmental time.

To achieve the purpose of this study we established the following null

hypotheses:

1)Infants with and without SB will demonstrate the same total number of
leg movements in each condition.

2) Infants with and without SB will demonstrate the same total number of
leg movements over developmental time.

3) The infants will demonstrate bilateral sensitivity to the weightings by
increasing the proportion leg movements between the unweighted limb relative to
that of their weighted limb.

4) There will be no significant relationship between any of the
anthropometric measures and the total frequency of leg movements generated.

Methods

Participants:

Prior to participant recruitment, Saint Catherine University Institutional

Review Board approval was obtained. The participants were four infants with

lumbar or sacral SB who ranged in age from five to 11 months at entry into the

study, none of whom were premature. We selected this age range because there

is a relative lack of information about the leg movements of infants with SB

beyond seven months of age. This is also a range in which it is easier to observe

babies' leg movements secondary to the babies not being able to walk during this

time period. We limited our study to infants with SB due to the extensive body of

knowledge that currently exists regarding the spontaneous leg movements of TD babies.

The faculty advisor for this study recruited the participants through the state of Minnesota Spina Bifida Association and the SB clinic of a large metropolitan hospital located in the upper Midwest. All participants earned a $10.00 per visit participation incentive. An initial phone call was made to the infants' families, which included an explanation of the purpose, risks and benefits, and the time commitment of the study as well as scheduling the initial visit. Table 4 in the results section of our paper presents a summary of characteristics for each of the participants.

Movement Data Collection

All data were collected in each baby's home at a time when the parent or caregiver reported that their infant was generally alert and active. After meeting with the parent(s) and baby, informed consent was obtained at the first visit. Following this and at each subsequent visit, the baby's clothes were removed from his/her legs to allow for maximal freedom of movement. Three anthropometric measures were collected at the beginning of each session. These included the baby's weight, calf length and circumference which were entered into an Excel spread sheet to calculate the baby's estimated calf mass. This approach was based on Schneider and Zernicky's previous research that showed calf mass can be accurately estimated based on body mass, calf length, and calf circumference.[36] A complete explanation of the 11 the anthropometric measures are summarized in Table 3.

Table 3.

Anthropometric and Range of Motion Measures

Anthropometric Variable	Measurement Technique	
Total Body Length	Baby is supine, looking up, body aligned with ankle dorsiflexed to 90 degrees. Measurement is taken from the top of the head to the bottom of the foot and recorded in inches to the nearest 1/16th of an inch.	
Thigh Length	The distance between the greater trochanter and the lateral condyle of the femur with the leg extended and the ankle dorsiflexed to 90 degrees	
Calf Length	The distance between the lateral condyle of the femur and lateral malleolus with the leg extended and the ankle dorsiflexed to 90 degrees	
	NOTE: Both leg length measures recorded in centimeters and measured to the nearest millimeter.	
Weight	Measured in pounds and ounces to the nearest ounce.	
Thigh Circumference	The circumference of the thigh measured at the midpoint of the segment length as measured above	
Calf Circumference	The circumference of the calf measured at the midpoint of the segment length as measured above	
	NOTE: Both circumference measures recorded in centimeters and measured to the nearest millimeter and taken on the right leg.	
Medial Thigh Skinfold	The skinfold is vertical and taken at the same level as the thigh circumference on the medial surface with the thigh extended	
Medial Calf Skinfold	The skinfold is vertical and taken at the same level as the calf circumference on the medial surface with the calf and knee extended	
	NOTE: Both skinfold measures recorded in millimeters and measured to the nearest .5mm and taken on the right leg.	
Ankle Plantarflexion/Dorsiflexion	The infant is supine with the leg extended at the knee as the ankle is moved into plantarflexion and dorsiflexion	
	NOTE: Both range of motion measures recorded to the nearest degree and taken on the right lower extremity.	

Next, small fishing weights that were equal to 25%, 50%, 75%, and 100% of the infant's estimated calf mass were placed in four small wrist sweat bands, respectively. Small reflective markers were then placed on the head of the first metatarsal of each foot. Then, the baby was placed in a specially designed infant seat designed to facilitate leg movements. The position was based on Chapman's earlier work in which he reported that a more upright posture resulted in more frequent leg movements by infants with SB compared to when they were supine or seated in a conventional infant seat.[7,23,26] The specially designed infant seat, as shown in Figure X, positioned the infant's trunk at an angle of 37 degrees from vertical. It provided firm support, but, with the exception of the head support area, did not constrain lateral arm or leg movements. The base of the seat was 33 cm high, thus preventing the infant's feet from touching the support surface. An elastic cloth was positioned around the infant's chest and fastened with Velcro behind the back support to stabilize the infant's trunk. During testing, the parent or faculty advisor was seated near the infant's side for each trial and interacted with the infant visually and socially to encourage activity. While seated in the infant seat, the baby's leg movements were videotaped at 30 frames per second for one minute per condition using a Sony HD Handycam video camera that was located perpendicular to and two meters from the infant.

The infant was videotaped in each of the following conditions: Baseline (no weight added to either leg), then with 25%, 50%, 75%, and 100% of his/her estimated calf mass on one leg. The order of leg, right and left was randomized per coin toss and the order of weightings, 25%, 50%, 75%, and 100% were

randomized per a computerized random number table for each baby and visit. This resulted in a total of 9 minutes of videotaped data for each visit. Each infant received a brief rest period between leg conditions. Data were collected one time per month for three consecutive months for each baby.

Before videotaping each infant, we videotaped a 1.0-meter long calibration rod that was placed within the space to be occupied by the infant, parallel to the floor for one minute. Test sessions were videotaped with a Sony HD Handycam video camera that allowed for two-dimensional position-time data to be collected during all trials. Sampling rate was 30 hertz. The camera was mounted on a tripod and positioned up to a maximum of two meters from and perpendicular to the infants' feet with the optical axis of the camera lens set at an angle of 22.5 degrees from the floor. This resulted in a camera height of 82 cm when the camera was two meters from the feet of the baby. Two-dimensional (2-D) data were collected rather than three-dimensional (3-D) data because our primary interest was in describing how often infants with SB move their legs rather, than, for example torque values of infants' leg movements, which would have required a 3-D technique.

Anthropometric Data Collection

After the first set of movement test trials were completed, the remaining anthropometric measurements were taken by the faculty advisor so that we could examine if the selected physical characteristics were related to the amount of leg movement observed. The remaining anthropometric measures included: total

body length; thigh length and circumference; medial thigh and calf skinfolds; and ankle plantar- and dorsi-flexion. All length measures were taken on the right leg. We anticipated that infants who gained more weight over time or who had larger skinfold measures and/or greater leg circumferences would move their legs less often than leaner babies who have thinner legs. We also expected that infants with longer leg segments would move their legs less often than infants with relatively shorter leg segments. The range of motion variables were measured to determine if infants with increased joint laxity moved their legs more frequently than babies with less mobile joints. Each of these measures were taken on each test day. See Table 3 for a complete description of the anthropometric measurement technique.

Data Reduction

The videotaped data for each baby's set of trials was behavior coded frame by frame to determine the frequency of leg movements for each leg. A leg movement was defined as "when a movement started then stopped or a change in direction occurred".[7] (pg 18.) For example, if a baby moved his or her leg medially then reversed direction and moved the leg laterally, the first movement ended and the second began at the point of change in direction. Each student researcher achieved a percent agreement of $\geq$ .85 with an expert rater prior to behavior coding any of the videotaped data. After tabulating the observed frequencies for each trial, the number of leg movements generated per minute was calculated.

Data Analysis

The data were analyzed by calculating the average per minute frequency of total leg movements in each condition at each age and for each leg. The per minute average for each baby's leg movements in each condition provided the data needed to determine if there was a statistical difference between conditions, percentage of left and right leg movements, and over developmental time (three months). Multivariate MANOVA procedures for condition and age were implemented via SPSS – Version 19 software to confirm or deny statistical differences. Post hoc analyses included Tukey post-hoc test(s) and inspection of the means for significant main effects. Pearson product-moment correlation coefficients were calculated between the anthropometric measures and TLMs generated in BL as well as between the left and right legs in each condition and over time to determine the strength of the relationship between how often the left and right legs moved, respectively.

Results

Description of Participants

Four participants completed this study (100% completion rate). All were females. Each infant had a lumbar or sacral level spinal lesion. None were taking medications on a consistent basis, but each had at least one episode of antibiotic therapy secondary to either upper respiratory or urinary tract infections during the course of this study. A description of the birth history and individual characteristics of the 4 infants is presented in Table 4.

Table 4.

Participant Characteristics

Characteristic	Baby 1	Baby 2	Baby 3	Baby 4
Age at entry (mo)	11	5	9	7
Gestation age at birth (weeks)	39	37	39	36
Delivery method	Vaginal	C-section (planned)	Vaginal	C-section (planned)
Lesion level	S1	L1-2	L4-5	L4-5
Medication	None	None	None	None
Co-morbidities/ Past medical history	Tethered cord release	VP shunt, B hip dysplsia, CPAP at night	VP shunt, Chiari II malformation with paralyzed vocal chords, tracheostomy, G-tube, B hip dysplasia	B club feet, 4 VP shunts, 1 ventricular – artial shunt, tracheostmy, Chiari II malformation with paralyzed vocal chords, G-tube, Pierre Robin Syndrome, cleft palate, B hip dysplasia, missing B patella, limited knee flexion to ~20° B

VP=ventriculoperitoneal, B=bilateral, CPAP= continuous positive airway pressu

Total Leg Movements and Percentage of Left and Right Leg Movements

Our first null hypothesis stated that the infants would demonstrate the same total number of leg movements in each condition. To determine the veracity of this prediction we conducted a multivariate analysis of the total number of leg movements and the percentage of the total leg movements each leg contributed. Each weighted condition including baseline and age were the

independent variables. The average number of total leg movements (TLM) generated in one minute and the percentage of left and right leg movements were the dependent variables.

We found a significant main effect for the condition variable and TLMs produced per minute {F (3,24) = 2.781, p = .009}. There was not a significant difference between conditions for the percentage of left and right leg movements {for percent left F(3,24) = .701, p = .690; for percent right F(3,24) = .689, p = .700)}.

A Tukey post hoc analysis was then completed to establish where the significant difference(s) occurred between conditions. This analysis showed a significant difference in TLM each minute between the baseline (BL) condition and each of the following weighted conditions: 25% left weighted (LW), 25% right weighted (RW), 50% LW, 50% RW, and 75% LW. There was not a significant difference in the number of TLM generated by these infants between the BL trial and these weighted conditions: 75% RW, 100% LW, and 100% RW. This lack of significance is likely due to our small sample size and the relative Significant Differences in TLMs Generated Between BL and Weighted Conditions variability demonstrated by this group of infants. Table 5 shows the significant level for each weighted condition as compared to BL.

Table 5.

Significant Differences in TLMs Generated Between BL and Weighted Conditions

Condition	p Value (significant*)
25 % LW	.010*
25% RW	.030*
50% LW	.005*
50% RW	.033*
75% LW	.027*
75% RW	.106
100% LW	.279
100% RW	.066

Next, we inspected the means for each trial to determine in which condition the infants generated the most and fewest TLM per minute. This revealed that there was a significant increase in TLM in each of the weighted conditions as compared to BL. In other words, the babies moved their legs significantly less often when they had no weights attached to one of their legs. These results are illustrated in Figures 1-4 which depict the means and standard deviations for TLMs in each condition for the trials when the left leg is weighted; TLMs when the right leg is weighted; the percentage of left and right leg movements when the left leg is weighted; and the percentage of left and right leg movements when the right leg is weighted, respectively.

Figure 1. Bar graph showing average total leg movements in each left leg weighted condition.
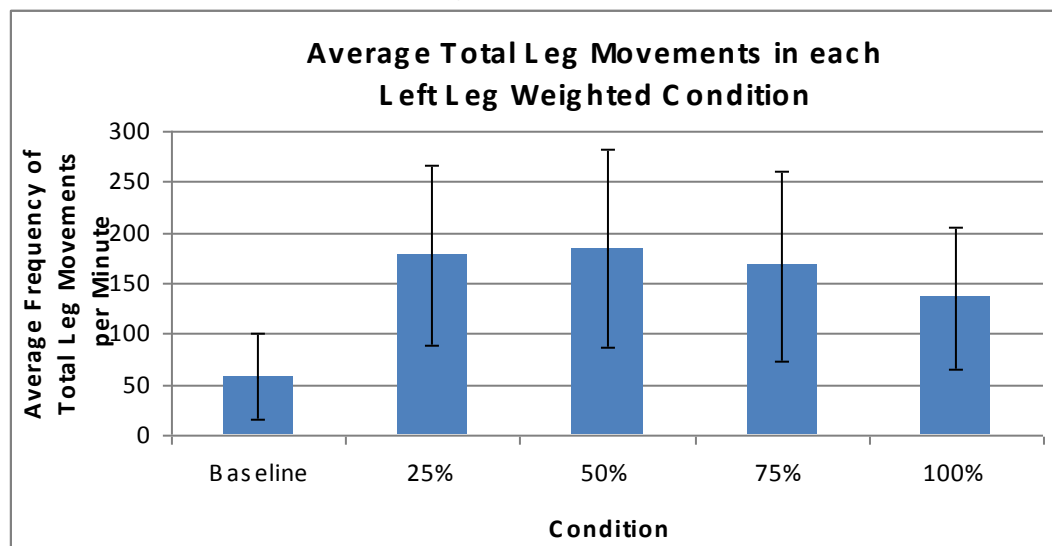

Figure 1. Average frequency of total leg movements (movements/min) in each left leg weighted condition over developmental time. Error bars represent one standard deviation.

Figure 2. Bar graph showing average total leg movements in each right leg weighted condition.

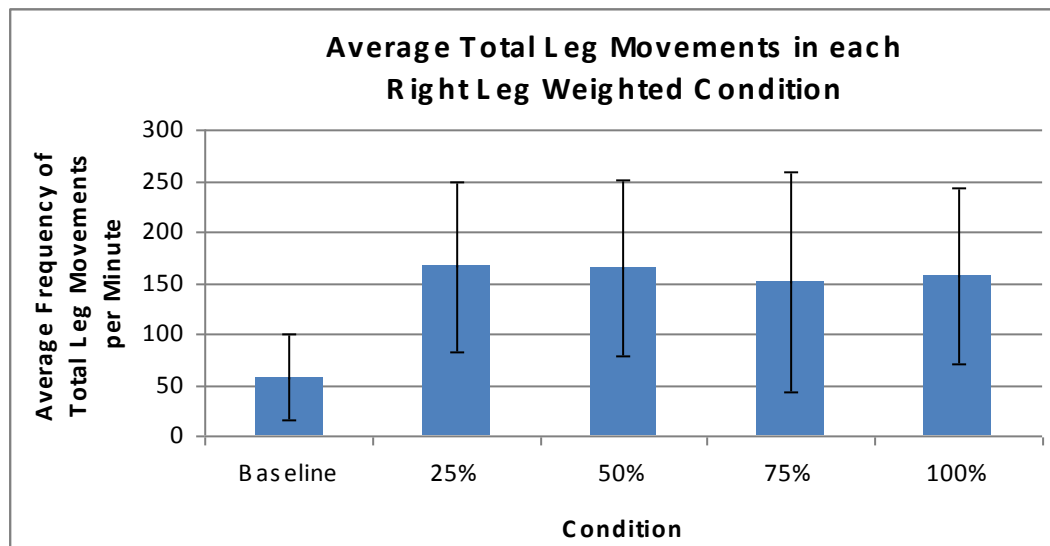**Average Total Leg Movements in each Right Leg Weighted Condition**

Figure 2. Average frequency of total leg movements (movements/min) in each right leg weighted condition over developmental time. Error bars represent one standard deviation.

Figure 3. Bar graph showing the average percentage of left and right leg movements in each left leg weighted condition.

Figure 3. Average percentage of left and right leg movements in each left leg weighted condition over developmental time. Error bars represent one standard deviation.

Figure 4. Bar graph showing the average percentage of left and right leg movements in each right leg weighted condition.
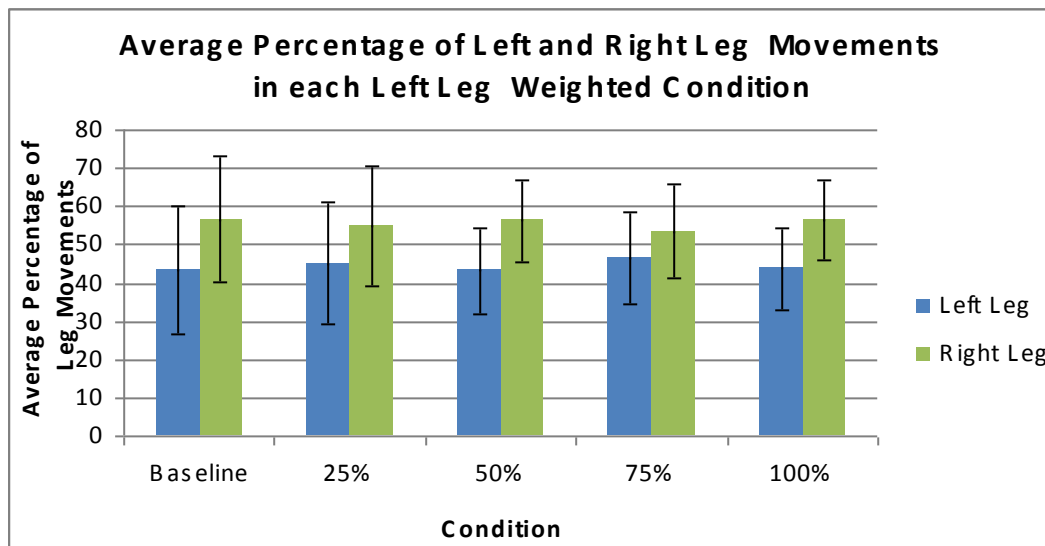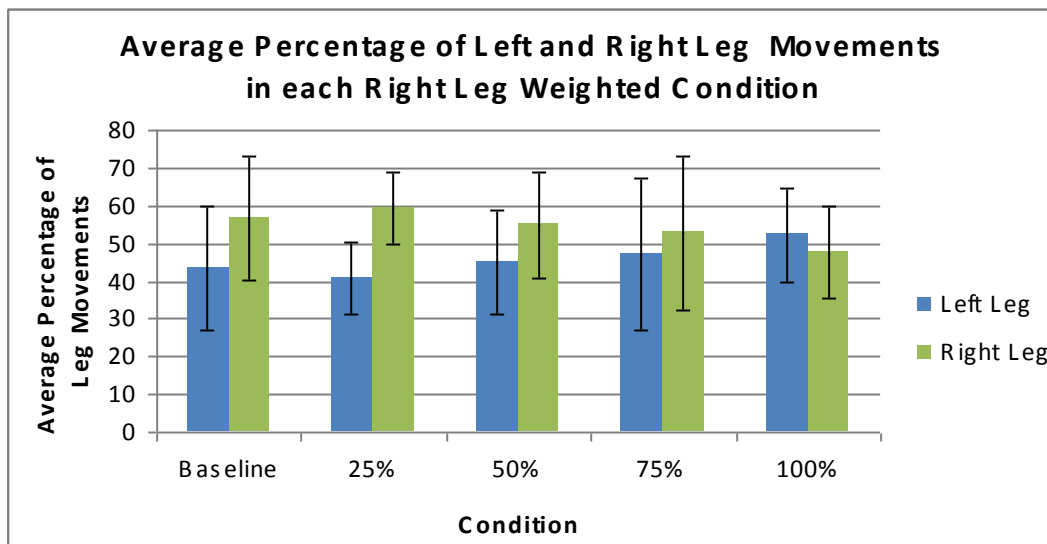

Figure 4. Average percentage of left and right leg movements in each right leg weighted condition over developmental time. Error bars represent one standard deviation.

In our second hypothesis we predicted that the infants would demonstrate the same number of TLM over developmental time. Our statistical analysis did not support this null hypothesis. Rather, there was a significant main effect for age with respect to the TLM produced by these infants per minute as well as the percentage of left and right leg movements demonstrated in each condition at each age. The specific F values are as follows: $F(2,6) = 13.305$, $p = .00$ for TLM generated over developmental time, $F(2,6) = 7.122$, $p = .001$ for percentage of left leg movements over time; and $F(2,6) = 6.953$, $p = .002$ for percentage right leg movements over time.

Then, we submitted this data to a Tukey post hoc analysis to determine when in development the significant differences occurred for TLM and the percentage left and right leg movements. This analysis confirmed a significant

difference in TLM produced by the babies when they were 8.25, 9.25, and 10.5

months old. Table 6 shows the significant levels in TLM between the

developmental ages. Inspection of the means showed a significant increase in

TLM generated between when the infants were 8.25 and 9.25 months of age and

a significant decrease in TLM when they were 10.5 months old as compared to

the two youngest ages.

Table 6.
Differences in TLM, Percent Left, and Percent Right Between Ages

Ages	TLM Significance	% Left Significance	% Right Significance
8.25 to 9.25	.042*	.140	.139
8.25 to 10.5	.023*	.001*	.001*
9.25 to 10.5	.000*	.159	.174

For the percentage of left and right leg movement data, the Tukey post

hoc analysis highlighted a significant difference between when the infants were

8.25 and 10.5 months old, respectively. Table 6 also shows the significant levels

in percent left and percent right leg movements between the developmental ages

as well. Inspection of the means indicated that there was a significant decrease

in the percentage of left leg movements and a significant increase in the

percentage of right leg movements between the ages of 8.25 and 10.5 months.

Figures 5-6 illustrate the means and standard deviations of the TLM and the

percentage of left and right leg movements at each age, respectively. There was

not a significant interaction effect between condition and developmental age for

TLM $\{F (16,48) = .622, p = .822\}$, percentage of left leg movements $\{F (16,48) =$

.697, *p* = .789}, and percentage of right leg movements {F (16,48) = .713, *p* = .773}.

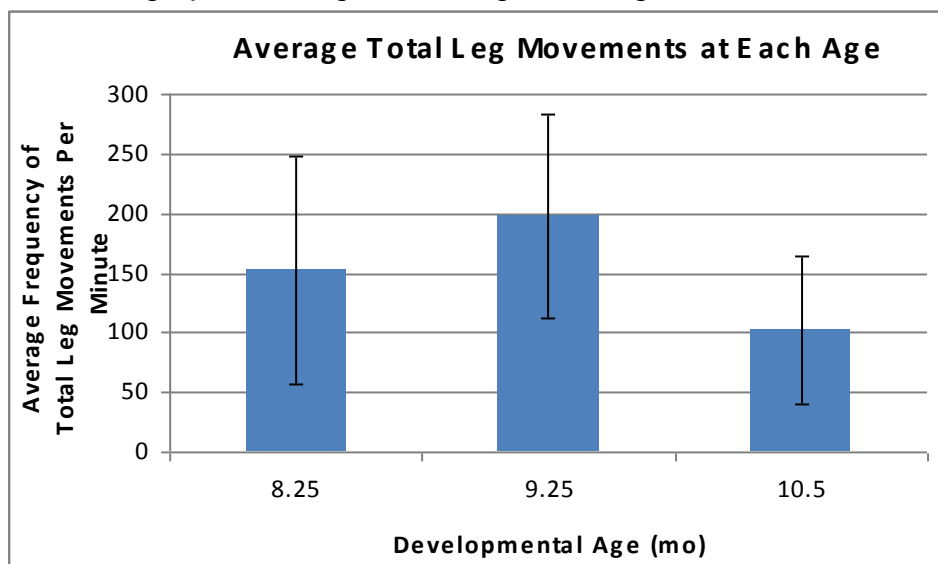Figure 5. Bar graph showing the average total leg movements at each age.

Figure 5. Average frequency of total leg movements (movements/min) at each age across all conditions. Error bars represent one standard deviation.

Figure 6. Bar graph showing the average percentage of left and right leg movements at each age.
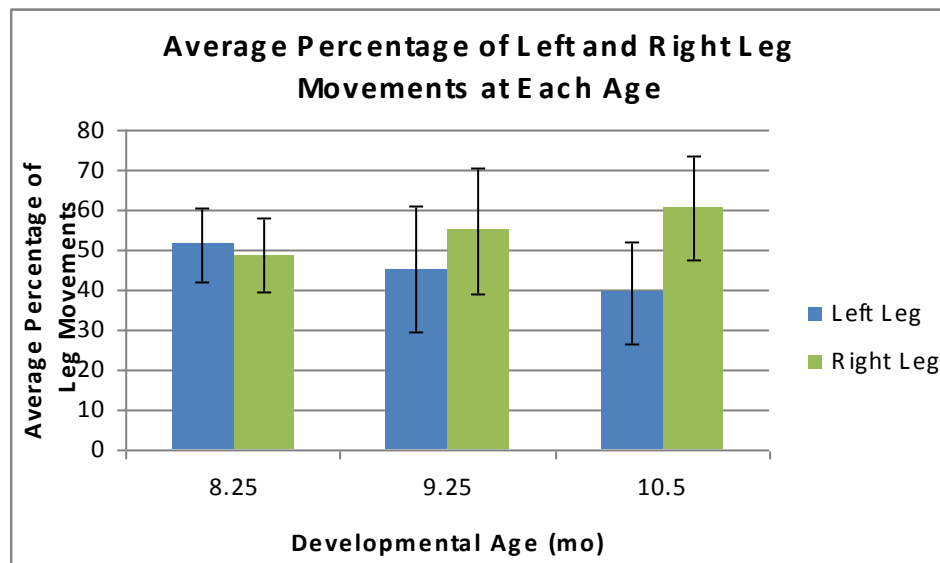

Figure 6. Average percentage of left and right leg movements at each age across all conditions. Error bars represent one standard deviation.

Leg Weightings and Interlimb Coupling

In our third hypothesis we predicted that this group of infants would, at some level of weighting, 25%, 50%, 75%, or 100%, 'un-couple' their legs and begin to move the un-weighted leg more often than the weighted leg. To test this hypothesis, we calculated Pearson product-moment correlations between the right and left leg movements across condition and over time to determine which, if any, demonstrated a significant ($p < .05$) relationship with each other. This analysis revealed a significant correlation between the left and right leg movements ($r = .282$, $p = .003$) across conditions and over developmental time. This indicates that when one leg moved so did the other leg. This result suggests that this small group of infants did not 'un-couple' their legs at any of the given

weightings and that they maintained a level of inter-limb coupling that was similaa

to the BL trials as they got older.

Relationship between Anthropometric Variables and TLMs

In our final hypothesis, we expected that there would not be a significant

relationship between any of the anthropometric measures and the TLM

generated in each condition at each age. The anthropometric data that we

collected included: length variables (height, thigh and calf length); mass variables

(weight, thigh and calf circumference, thigh and calf skinfold); and range of

motion variables (ankle plantarflexion and ankle dorsiflexion). The means and

standard deviations of the anthropometric variables by month are presented in

Table 7-9.

Table 7.
Length Variables: Means and Standard Deviations by Month

Age (mo.)	Height (in)	Thigh Length (cm)	Calf Length (cm)
8.25	26.0 $\pm$ 3.5	11.9 $\pm$ 2.9	12.2 $\pm$ 2.2
9.25	26.5 $\pm$ 3.6	12.1 $\pm$ 2.1	12.3 $\pm$ 6.6
10.5	27.4 $\pm$ 3.7	12.6 $\pm$ 2.8	12.6 $\pm$ 2.5

Table 8.
Mass Variables: Means and Standard Deviations by Month

Age (mo.)	Weight (lbs)	Thigh Circumference (cm)	Thigh Skinfold (cm)	Calf Circumference (cm)	Calf Skinfold (cm)
8.25	14.6 $\pm$ 4.5	20 $\pm$ 5.6	20.4 $\pm$ 13.1	14.4 $\pm$ 3.5	20.3 $\pm$ 9.1
9.25	16.2 $\pm$ 5.1	20.2 $\pm$ 4.3	19.4 $\pm$ 10.9	15.2 $\pm$ 3.8	19.6 $\pm$ 9.9
10.5	16.1 $\pm$ 5.1	21.3 $\pm$ 5.2	20.2 $\pm$ 9.7	15.5 $\pm$ 4.1	18.5 $\pm$ 7.4

Table 9.
Range of Motion Variables: Means and Standard Deviations by Month

Age (mo.)	Plantarflexion (deg)	Dorsiflexion (deg)
8.25	53.8 $\pm$ 42	18.7 $\pm$ 19
9.25	46.3 $\pm$ 31	27.5 $\pm$ 23
10.5	65 $\pm$ 21	28 $\pm$ 21

The anthropometric variables grouped by type of variable, i.e. length, mass, and range of motion and their relationship with TLMs are presented in Tables 10-12. We calculated Pearson product-moment correlations between the anthropometric measures taken over developmental time to determine which, if any, demonstrated a significant ($p < .05$) relationship with the TLMs generated during the BL condition. We chose to assess the strength of the relationship between the anthropometric variables with the TLMs at BL because previous research has shown only limited to moderate correlations between anthropometric data and frequency of movement data as well as the small sample size included in this pilot project.

None of the length variables demonstrated a significant relationship with TLMs (See Table 10). The only significant correlation between any of the mass variables and TLMs produced during the BL condition were thigh skinfold ($r = -.609$, $p = .036$) and calf skinfold ($r = -.599$, $p = .039$). These moderate negative correlations indicate that infants with larger skinfolds tended to move their legs less often than babies with smaller skinfolds (See Table 11). It should be noted that with a larger sample it is likely that the thigh and calf circumference variables would also reach significance. As seen in Table 12, ankle plantarflexion was the

only range of motion variable that demonstrated a significant relationship with

TLM generated during BL trials (r = -.656, p = .021). This moderate negative

correlation value suggests that babies with more passive range of motion moved

their legs less often compared to infants with smaller amounts of passive range

of motion.

Table 10.

Pearson Correlation Values between Length Variables and TLM at BL

	Height	Thigh Length	Calf Length
Correlation	-.198	-.300	-.421
Sig. (2-tailed)	.537	.344	.173

Table 11.
Pearson Correlation Values between Mass Variables and TLM at BL

	Weight	Thigh Circumference	Thigh Skinfold	Calf Circumference	Calf Skinfold
Correlation	-.470	-.551	-.609*	-.574	-.599*
Sig. (2-tailed)	.123	.064	.036	.051	.039

Table 12.
Pearson Correlation Values between Range of Motion Variables and TLM at BL

	Plantarflexion	Dorsiflexion
Correlation	-.656*	-.522
Sig. (2-tailed)	.021	.082

Discussion

Our goal for this study was to build upon the existing physical therapy literature regarding how infants with SB use sensorimotor information as they learn to produce coordinated leg movements over developmental time. Our specific aim was to examine how infants with lumbar or sacral SB respond when small amounts of weight are added to one of their legs. As a result, the purpose of this study was to determine if infants with lumbar or sacral SB, between five and 11 months of age at entry into the study, move their legs more or less often when they have 25%, 50%, 75% and 100% of their calf mass added to one of their legs while they were seated in an infant seat designed to facilitate leg movements over three months of developmental time.

In our first hypothesis we predicted that the infants would demonstrate the same number of total leg movements in each condition. Based on previous research we anticipated that they would accomplish this by decreasing how often they moved the weighted leg and increasing how often they moved the un-weighted leg. Our results, in contrast to previous literature, show that the babies moved their legs significantly more often in the weighted conditions compared to the BL trials and did not change the percentage of leg movements, i.e. weighted versus un-weighted, generated in each condition.

This differs from previously published literature that documents how infants with and without Ds responded to this type of paradigm. Ulrich et al showed that TD infants and infants with Ds maintained their overall frequency of leg movements in each weighted condition (25%, 50% and 100%) by increasing

the frequency of their un-weighted limb and decreasing how often they moved their weighted leg. The babies in our study generated significantly more leg movements when weight was attached to either of their legs, but did not adjust how often the weighted verus the un-weighted leg moved. Thus, this small sample of infants with SB increased their overall frequency of leg movements. And, they did not move their weighted leg less compared to their un-weighted leg. These findings seem to suggest that infants with lumbar or sacral SB may process sensory information differently compared to TD infants and/or infants with Ds. This processing difference may contribute to the delays they experience in learning to produce the functional leg movements needed to crawl, pull to a stand, and walk.

In spite of the fact that this small group of infants with SB did not respond to this perturbation like TD infants and infants with Ds we are encouraged by the fact that all four infants increased their leg movements in each weighted condition. As noted earlier, infants with SB tend to move their legs less often than TD babies. (Chapman 99) Thus, an intervention of this type may be used to help babies with SB move their legs more often compared to when they have less or different sensory information available to them. Increased leg movements should lead to increased muscle strength and endurance as well as strengthened neural connections that support their ability to move their legs (Edelman). This type of effect may, over time, lead to their ability to produce functional leg movements earlier in life than is currently reported in the literature.

Based on the existing literature we anticipated that the infants in our sample would demonstrate the same number of total leg movements over developmental time.[7,23,26] Our statistical analysis did not support this expectation. Rather, we found that this set of infants had a significant increase in frequency of leg movements between 8.25 and 9.25 months of age with the most increase at 9.25 months. We then saw a significant decrease in frequency of leg movements at 10.5 months of age. We also found that there was a significant difference between when the infants were between 8.25 months and 9.25 months respectively. The interpretation of our results is challenging in that there have been no previous longitudinal studies utilizing this paradigm. This data set is inconsistent with Chapman's earlier work. However, his previous studies were designed to examine the frequency of leg movements when infants with SB were in different positions rather than manipulating sensory information directly available to the babies. In light of this there is a need for additional research in this area.

Previous research has shown that infants with and without Ds will demonstrate bilateral sensitivity to the weightings by increasing the proportion of leg movements between the unweighted limbs relative to that of their weighted limb. As a result, we predicted that our group of infants would also increase the proportion of leg movements between the unweighted limbs relative to that of their weighted limb. Our results showed a significant correlation between the left and right leg movements across conditions and over developmental time. These findings suggest that at each weighting and over developmental time the babies

maintained their interlimb coupling. This is unlike previous data with TD infants and infants with Ds that showed a decrease in leg movements of the weighted limb and increased movements of the unweighted limb. What this suggests for infants with SB is there is a decrease in sensitivity to the different leg weighting conditions compared to infants who participated in previous research studies. This implies that infants with SB in general may move less frequently secondary to the lack of reward they experience with leg movement. Diminished sensitivity and less frequency of movement is detrimental to their reaching gross motor milestones compared to their TD peers.

Our final hypothesis was that there would be no significant relationship between any of the anthropometric measures and the total frequency of leg movements generated in the BL trials. Our results revealed a significant negative correlation between calf and thigh skinfold values and how often the babies moved their legs. This suggests that babies with larger skinfold measures moved their legs less often in BL compared to infants with smaller skinfold measures. There was also a significant negative correlation between plantar-flexion and the frequency with which this group of infants moved their legs in BL. This suggests that babies with more joint laxity do not move their legs as often as infants with less passive plantar-flexion of the foot at the ankle. Based on these results, it is possible that infants with SB potentially lack proprioceptive input from their joints and supports Chapman's perceptions regarding the disrupted sensiormotor pathways experienced by infants with SB.[7]

Conclusion

Our goal for this pilot study was to build upon the existing literature regarding infants with SB and how they respond to environmental changes. Using a 2-D approach at 30 frames per second we found that by simply adding small weights to one of their legs caused an increase in how often they moved their legs. This shows that babies with SB are sensitive to changes in afferent input, but because this group of babies did not show the ability to uncouple their legs like TD babies and babies with Ds it seems that they use this type of information differently compared to previously studied babies. Instead, this small group of infants with SB showed a strong tendency of moving their legs together regardless of the weighted condition. The results regarding the anthropometric data should be interpreted with caution in that our sample size was small which limits our ability to make inferences based upon this data. Future studies will be designed to examine if infants with SB who are seated in the specially designed seat generate more kicks when they have small weights added to one of their legs. We will also be working to determine if there is an optimal amount of weight that results in the highest number of leg movements.

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