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THE INFLUENCE OF AGE, POSITION, AND TIMING OF SURGICAL REPAIR ON THE KICKS OF INFANTS WITH SPINA BIFIDA

by Ann Engstrom Shannon Lucken Kayla Sis Sarah Wehrheim

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Date (April, 30, 2014)

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ABSTRACT

TITLE: The Influence of Age, Position, and Timing of Surgical Repair on the Kicks of Infants with Spina Bifida

BACKGROUND AND PURPOSE: We know little about the leg movements and kicks of infants with SB who have their spinal lesion repaired in-utero. Nor do we know how sensitive they are to the movement context in which they are placed. The purpose of this pilot study was to describe how often infants with SB who have had their spinal lesion repaired pre-natally move their legs and kick when they are in different postures.

SUBJECTS: Four infants with lumbar spina bifida. Two had pre-natal spinal surgery.

METHODS AND MATERIALS: Each infant was videotaped once a month for four months when they were supine and seated in a specially designed adjustable infant seat. Frame by frame behavior coding was used to identify leg movements and kicks. Anthropometric measures were collected to determine if these traits were related to how often they moved their legs or kicked.

ANALYSES: Due to the small sample size (n=4) descriptive statistics were used to verify trends in the number of leg movements and kicks generated in each position at each age. Correlations were calculated between the anthropometric measures and frequency of leg movements and kicks.

RESULTS: All four infants produced similar numbers of leg movements and kicks when supine and markedly increased how often they moved their legs and kicked when seated. The two infants who had in-utero surgery demonstrated more leg movements or kicks in each seated position at each age compared to the infants who had post-natal spinal surgery. Only thigh skinfold and plantarflexion were significantly correlated with leg movements or kicks (p=.05).

CONCLUSIONS: Infants with spina bifida who had their spinal lesion repaired in-utero move their legs and kick quite often. They appear to be sensitive to the movement context in which they are placed.

IMPLICATIONS: Therapists and parents should consider how the movement context influences the leg movements and kicks of infants with spina bifida.



The Influence of Age, Position, and Timing of Surgical Repair on the Kicks of Infants with Spina Bifida

> submitted by Ann Engstrom Shannon Lucken Kayla Sis Sarah Wehrheim

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

Primary Advisor David Chapme PT Php Date 4/30/14

Co-Advisor _____ Date____



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Introduction

There is limited literature in the field of physical therapy regarding the motor development of infants with spina bifida (SB). For this reason, our goal for this study is to expand the literature pertaining to infants with lumbar or sacral myelomeningocele (MM) by examining the influence age, position, and timing of the infant's spinal repair have on her motor development.

Spina bifida is the most common neural tube birth defect in the United States.¹ Between 1500 and 2000 infants are born with SB each year in the United States.¹ Spina bifida is characterized by malformations of the vertebral arches and protrusion of the spinal cord and associated nerve roots.² The closure of these arches typically occurs during the fourth week of embryogenesis but for infants with SB this process is disrupted.²

As summarized in Table 1 infants born with SB often experience motor, sensory, and cognitive impairments.² Common musculoskeletal impairments include scoliosis, clubfoot, and an increased risk for osteoporosis. Visual-perceptual deficits, cranial nerve palsies, and neurogenic bowel and bladder are common sensory impairments that impact infants born with SB. Babies and children with SB may also show signs of cognitive impairments and learning disabilities.



Motor Impairments	Sensory Impairments	Cognitive Impairments
• Musculoskeletal deformities	•Latex allergy	•Hydrocephalus
• Increased risk of osteoporosis	• Visuoperceptual deficits	•Language
• Motor paralysis	•Cranial nerve palsies	
•Obesity	•Skin breakdown	
•Upper limb discoordination	•Neurogenic bowel and bladder	
• Spasticity		

Table 1. Common Impairments Associated with Spina Bifida.

Currently, it is thought that SB develops as a result of one or more risk factors and is usually detected via sonogram, amniocentesis, or blood testing of the mother.^{2,3,4} For example, decreased folic acid intake has been linked to SB.^{2,3} The documentation of the relationship between this nutritional deficit and the development of SB has resulted in the United States Public Health Service's recommendation for women who are pregnant, or could become pregnant, to consume 400 micrograms of folic acid daily. The use of anti-convulsant medications during pregnancy is also commonly linked to SB.³ These, as well as other risk factors, such as race (there is a greater prevalence among whites and hispanics), family history, and maternal hyperthermia have shown some correlation with SB, but none have been identified as a direct cause.³

There are multiple types of SB that fall in the broad category of myelodysplasia or defect in the spinal column.^{2,5} Spina bifida occulta is the mildest form of SB.



Individuals with this form of SB are often asymptomatic. They may present with a patch of hair or dimple along the back where the spinal lesion is located.^{2,5} With this type of SB, the vertebral column is not completely closed, but it is not open enough to allow for the spinal cord to protrude dorsally.

Spina bifida aperta results in open and visible lesions and can further be delineated into meningocele and myelomeningocele (MM).^{2,5} The gap in the vertebral arches of meningocele lesions is large enough for the meninges to protrude, but nervous system tissues are not exposed. Individuals with this type of SB may also be asymptomatic or may demonstrate the signs and symptoms associated with more complex neural tube defects.^{2,5}

The most serious type of SB is myelomeningocele. This form of SB is caused by an incomplete closure of the posterior vertebral arches.^{2,5} In this situation, the meninges and nerve roots protrude dorsally and can be seen and felt along the individual's spine. These lesions can occur anywhere along the spinal column, but nearly 75% are located in the lumbar and sacral region of the spine, especially in males.^{3,5} Figure 1 below illustrates all three forms of SB.¹





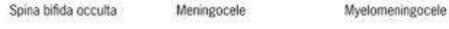


Figure 1. The three forms of Spina Bifida.ab

Most babies with SB are delivered via cesarean section to reduce the amount of trauma to exposed tissues associated with a vaginal birth. They then have surgery to repair the lesion within 24-48 hours after birth.^{3, 6} However, a relatively new technique of performing this surgery in-utero between 19 and 25 weeks gestation has been introduced.⁶ The motive for offering this pre-natal surgery is to reduce the need for a ventricular-peritoneal (VP) shunt, reverse hindbrain herniation, and enhance motor development.⁶ If the mother and fetus are deemed appropriate, the mother undergoes general anesthesia, which anesthetizes the fetus as well. An incision is then made along the abdomen and the uterus so that the fetus' back can be exposed. As with post-natal surgery, a pediatric neurosurgeon then removes the sac and places a skin flap over the opening. In addition to surgery remove the sac, many infants and children who are not operated on in-utero receive a VP shunt to assist with the drainage of cerebrospinal fluid that would otherwise build up and could cause hydrocephalus.



The location of the lesion and whether or not a VP shunt is placed are related to the amount and severity of impairments seen. Compared to lower level lumbar or sacral lesions, individuals with higher-level lesions experience greater levels of impairment.^{2,3} Due to the frequency of sensorimotor impairments, infants with SB often experience delays in achieving motor milestones compared to typically developing (TD) infants. These developmental markers for TD and infants/children with SB are displayed in Table 2.^{2,7}

Developmental Milestone	TD Infants	Infants with SB	
Head Control	3 months	+	
Rolling	4 months	+	
Sitting	6-7 months	1-2 years	
Crawling	7-11 month	1-2 years	
Standing	9-13 months	3 years	
Walking	12-14 months	3-7 years	

Table 2. Developmental Milestones.

+NOTE: Ages for these milestones are not documented in the literature.

Multiple theories have been used by developmentalists and therapists to describe when and explain how infants and children gain new motor skills. In particular, neuromaturation theory and ecological psychology have been used to describe when infants and children demonstrate selected motor milestones, like sitting, pulling to a stand, and walking. Proponents of the neuro-maturation approach believe that central nervous system (CNS) maturation, i.e. inhibition of primitive reflexes 'drives' typical motor development. They also believe that there is a certain pre-determined order of skill



acquisition that infants and children go through as they gain control over their developing bodies, e.g. they roll over before learning to sit, sit prior to pulling to a stand, crawl before walking. Finally, supporters of the neuro-maturation approach argue that motor development occurs from head to toe and proximal to distal.^{8,9}

Alternatively, ecological psychologists posit that based on the developing infant's understanding of what their environment 'affords' them to do they will demonstrate selected motor skills within a particular setting.⁷ This approach attempts to explain motor development by accounting for the interaction between the mover, their environment, and most importantly, the mover's perceptions of what the environment will allow them to do.⁷

The neuro-maturation approach has clearly contributed to our understanding of when most children demonstrate specific motor skills and provides the basis for a majority of age related standardized tests of gross motor development. Ecological psychologists broadened this work by encouraging parents, therapists and other providers to consider how the environment and the mover's perceptions of the environment may be used to positively or negatively impact how a given child moves. Both lack the ability to fully describe the motor behaviors infants display as well as explain the underlying processes that influence how infants move in real time and acquire new motor skills over developmental time. Nor do they account for the individual differences that exist between and among children. For example, why do some children learn to walk after never crawling? And, how do some infants with a central system lesion learn to walk in spite of having limited or no sensory information available to them from their legs? As a result,



researchers, developmentalists, and therapists have begun to use principles from dynamic systems theory (DST) to understand and then explain the developmental process that infants and children with and without disabilities go through as they learn to move their bodies in a coordinated manner.^{3,7, 10-13}

Advocates of the DST approach posit that the motor skills infants and children display in real time and develop over time are the result of self-organization as they encounter movement tasks within a given environment.^{3,6} Self-organization means that the infant organizes her multiple sub-systems, such as her muscles, joint receptors, central nervous system, and previous movement experiences, in a way that enables her to generate a coordinated movement that meets the demands of the movement task in a given setting.

Dynamic systems theorists hypothesize that each sub-system has the potential to contribute equally to a given movement pattern at a particular point in time.^{3,12-13} It is important to note that proponents of DST, in contrast to neuro-maturationists, do not believe that movements occur as a result of hard-wired pre-existing neural templates.³ Instead, they advocate that the CNS is one of many equally contributing sub-systems that the baby or child relies as she works to generate coordinated movements in real time and over developmental time. Dynamic systems theorists also believe, like ecological psychologists, that the environment or movement context plays an important role in facilitating or inhibiting the motor skills displayed by infants and children.^{6, 7} This implies that if the environment changes, e.g. changes in the support surface or the task is modified, i.e. asking a child to run rather than walk, you would anticipate seeing a



different movement pattern. This movement may not be the one you expected, just a different movement pattern.

Thelen provides several examples of self-organization in her line of research that was dedicated to uncovering the changes that occur in infants as they gain new motor skills.¹⁴ In one of her first studies, Thelen questioned the validity of the assumption that neural inhibition is the driver of motor development when she held four week old infants who no longer demonstrated the infant stepping response in chest deep water.¹⁴ Her results showed that babies who no longer demonstrated the typical infant stepping response when they were held on a firm table top did in fact have the ability to generate multiple alternating steps. She discovered this by holding the babies in chest deep water on the same day that she held them on a firm table top. She suggested that by placing the infants in the buoyant water environment they were able to overcome the effect of gravity to produce consistent alternating steps. Thelen hypothesized that if the nervous system controlled infant movements then the babies should not have been able to produce alternating steps in any environment.¹⁴

A second example from Thelen's work involves supporting babies on a small motorized treadmill.¹¹ She found that six to seven month old typically developing infants who had otherwise shown no ability to produce alternating steps were able to generate alternating steps when they were held on the infant sized treadmill.¹¹

Thelen et al provided another example of self-organization through her research with seven month old babies and a split-belt treadmill.¹² None of these infants were ambulatory, yet they produced alternating steps like her previous subjects when they



were held over the treadmill. However, by using a split belt treadmill, the researchers were able to double the speed of one belt while maintaining the original speed of the other belt to see if the infants would be able to adjust their steps to match the demands of the task. This team of researchers reported that the infants in this study adjusted their swing and stance phases to meet the demands of this unique environmental manipulation. Additionally, they found that when the belt speed was doubled, the infants increased the rate of their steps by 50%.¹²

Another example that supports the principle of self-organization comes from Chapman.^{13, 15} He demonstrated that the frequency of leg movements for infants with and without SB can be influenced by the movement context. Six TD babies and six infants with lumbar or sacral SB were examined over a fourth month period in a supine position, seated in a conventional infant seat, and in a specially designed infant seat. Both groups of infants, generated more leg movements and kicks when they were in the specially designed infant seat compared to when they were lying supine or seated in the conventional infant seat.¹⁰ He also examined the velocity and range of motion of the infants' leg movements and found that both groups of babies adjusted the velocity and range of motion of their leg movements depending on the context in which they were placed.

Self-organization, or the ability of the infant to adapt her movements to meet the demands of a task in a given environment, would not be possible without neuro-plasticity.^{3,6} Neuro-plasticity suggests that the relative strength or weakness of neural connections are influenced by how often the infant or child moves as well as the type of



movements she uses in real time and over developmental time. For example, a child who does not typically move her legs in alternation will not be able to strengthen the neural connections that support alternating leg movements. Alternatively, an infant who repeatedly reaches for objects with her left upper extremity will, in all likelihood, strengthen the neural pathways that support left-handed reaching. This implies that if the nervous systems of infants and children were not plastic, all movements in a given category of movement, e.g. alternating steps, would be the same regardless of the environment or other task demands. This concept provides additional support for the idea that movements are not hard-wired, but can be altered by collaboration between subsystems and changes in the environment or movement context. In Thelen's treadmill experiments, if neural plasticity was not available then the babies would have continued to generate alternating steps at a given rate regardless of the belt speed.¹⁴ Likewise, if infants' nervous systems did not have the ability to adapt and adjust the infants who participated in the split belt treadmill study would not have been able to adjust their stance and swing times to meet the demands of one treadmill belt going twice as fast as the other. Similarly, the infants with SB in Chapman's work would have shown the same types of leg movements in all three positions and would not have altered the velocity or amplitude of their leg movements when they were placed in different positions.

A second principle of dynamic systems theory is a rate-limiter. A rate limiter is a sub-system, either intrinsic or extrinsic to the mover, that prevents her from moving successfully, i.e. in a way that meets the demands of the task.^{3, 6} For example, rate limiters can be biomechanical factors, like the fat to muscle ratio of infants' legs,



diminished strength or a lack of active range of motion; neural factors, such as reduced or absent proprioceptive information, a CNS lesion; or environmental constraints, e.g. the support surface, ambient lighting in the environment.^{3, 13, 15}

In contrast to a rate limiter, a control parameter is a component of the system that acts as a primary agent of change, that is, one that enables the mover to demonstrate a new or more effective type of movement pattern.¹⁰ Control parameters are often rate-limiters that have been addressed through therapy and other movement experiences. For example, improved strength and active range of motion may enable someone to move more effectively. Likewise, reducing someone's fear or pain may result in a more effective movement pattern. Finally, there are times when a simple environmental change through improved lighting or a firmer surface results in improved movement patterns.

Thelen et al's 'fish tank' study referenced above provides an example of rate limiters and control parameters.¹⁴ Initially, they assessed these infants by holding them on a firm surface in her laboratory and video-taped their ability to generate alternating steps due to the classic infant stepping response. She then told the parents to come back to the lab when their baby no longer showed this stepping response when they were held on a firm surface at home. On the return visit, she again held the infants on a table to confirm that they no longer demonstrated the infant stepping response. Once this was completed she held them in the fish tank and filmed their steps. She noted that in this case, the movement context could function as both a control parameter and rate-limiter. That is, when the infants were held on a table the environment seemed to constrain their ability to produce alternating steps. Alternatively, when held in a water environment the



babies were able to generate multiple alternating steps. Thelen also weighed each infant during each visit and measured the infants' thigh and calf skin-folds. She found that the infants who gained the most weight and who had the largest thigh and calf skin-fold measurements produced the fewest steps and showed less range of motion in their steps when they were held in the fish tank.¹⁴ Thus, illustrating that a rate-limiter may also be an intrinsic sub-system.

The ideas of self-organization, control parameters and rate limiters do not explicitly explain the development of the CNS nor its interaction with the musculoskeletal system.¹⁰ Thus, proponents of DST rely on the Theory of Neuronal Group Selection as put forth by Edelman to begin to understand and then explain how the infant's developing nervous system interacts with the baby's other sub-systems to contribute to the production of coordinated movements.

Edelman suggests that groups of neurons within the nervous system that support categories of movements, e.g. upper extremity reaching are strengthened throughout an infant's life through exposure to tasks or repetitions of similar behaviors.¹⁶⁻¹⁸ Therefore, the more repetitions of a given type of movement, e.g. knee kicks that an infant experiences results in stronger neural connections that support that type of movement and will likely increase the infant's ability to replicate that movement in the future. Since every infant, even monozygous twins, experiences life differently, their neuro-developmental processes will also be different. This is the idea, coupled with self-organization and neural plasticity that is behind Chapman's use of the specially designed infant seat versus a conventional infant seat.^{13, 15} A typical infant seat inhibits certain



motions because of how it is designed to provide maximum support of the baby's head, trunk, and lower extremities. Because it inhibits certain types of leg movements it can lead to weakened neural pathways for some of the key leg movements that provide the foundation for walking. In particular, conventional infant seats restrict the ability of infants with SB to generate knee and leg kicks as well as explore certain portions of their hip and knee joint arcs of motion. The specially designed infant seat, in contrast, is designed to allow for more freedom of the legs in order to strengthen neural pathways that support flexion and extension of the legs at the hips and knees and enables the baby to generate greater numbers of leg movements and kicks. The frequency of leg movements has been directly related to when infants with Down syndrome start walking as well as for typically developing infants.¹⁹

Alternating leg movements, characterized by flexion and extension of the legs at the hips and knees, have been observed in-utero as early as 16 weeks for both TD babies and those with SB.²⁰ Ultrasound technology has allowed researchers to observe the frequency and quality of leg movements in-utero and note that infants with thoracic, lumbar, and sacral SB are able to move their legs at the hips just as often as TD fetuses.²⁰ For instance, Sival et al have reported that at 18-39 weeks gestation fetuses with thoracic or lumbar SB lesions generate kicks with similar amplitude and speed compared with TD fetuses.²⁰ They also reported that, within the first week after birth, the babies with SB decrease the number, amplitude, and type of their leg movements.²⁰

Rademacher, Black, and Ulrich closely examined babies at one, three, and six months of age with lumbar or sacral SB.²¹ They compared the frequency of leg



movements when these babies were placed supine to the frequency of leg movements generated by TD babies of the same age and in the same position. They found that the infants with SB had a lower frequency of movement and smaller amplitude of movement in these conditions.²¹

Further research by Chapman supports this and provides a rationale for babies with SB to be placed in a specially designed infant seat.^{13, 15} A lack of sensory input received by the infants with SB compared to their TD counterparts due to their spinal lesions suggests the babies with SB may not get enough sensory feedback or sensory information that is of a high enough quality to provide the motivation to move their legs that would enable them to strengthen the neural pathways for the leg movements that lead to walking. Chapman's research has shown that infants with lumbar or sacral SB move their legs and kick more when placed in a specially designed infant seat than in a traditional infant seat.^{13, 15} He observed the babies from four to twelve months of age and also noted that these babies do not increase or decrease how often they move their legs or generate kicks over developmental time. These studies were limited by the fact that the specially designed infant seat was fixed at 45 degrees from horizontal.

Collectively, these studies do not, explain why infants with lumbar or sacral SB do not change how often they move their legs and kick over developmental time like typically developing babies. In light of this, we sought to answer the following questions: 1. Is there an optimal position that enables infants with lumbar or sacral SB to move their legs and kick more often as they get older?



2. Is there a relationship between their age and their position in space relative to the horizontal that facilitates their ability to move their legs and kick over developmental time?

3. Is there a significant relationship between how often they move their legs and how frequently they generate kicks?

Methods

Participants

The participants in this study were four infants diagnosed with lumbar SB who were, on average, 38.5 weeks old at entry into the study. Prior to data collection, IRB approval was obtained through St. Catherine University and Gillette Children's Specialty Healthcare. All infants were subsequently recruited though the SB clinic at Gillette Children's Hospital in St. Paul, MN. The parents of the potential participants were initially screened by hospital personnel and then contacted by the research advisor to determine interest in participating in the study. Those who were receptive received a verbal explanation of the purpose, risks, benefits, and time commitment of the study during this initial phone call. Verbal permission for the infants to participate was also obtained during this phone conversation.

During the first data collection session, the parent(s) provided written informed consent and supplied their child's past medical and surgical history, medications, orthopedic impairments, and level of spinal lesion (e.g. L4-L5). Each subject received a participation incentive of a \$10 Target gift card, per visit, for participating in this study. A summary of the individual infant characteristics are presented in Table 3.



Infant	Gender	Lesion Level	Medications	Surgical History	Timing of Spinal Repair	Orthopedic Impairments
1	Female	L5-S1	None	None	Post-Natal	None
2	Female	L4-5	Antibiotics Anti-Seizure Medications	V-P Shunt	Post-Natal	None
3	Male	L4-5	Vitamin D Drops	None	Pre-Natal	None
4	Female	L4-5	Antibiotics	None	Pre-Natal	None

Table 3. Infant Characteristics.

Movement Data Collection

Data collection for each infant occurred once a month for four consecutive months. The time of day for collecting the data was scheduled when the parent(s) suggested their infant would be alert and active. At the beginning of each data collection session, a 0.5 meter calibration rod was placed parallel to the floor in the space the baby would occupy during each trial. The calibration rod was videotaped for one minute. This enabled us to collect 2-D data, rather than 3-D data, as we were most interested in documenting the infant's frequency of leg movements and kicks. To allow for maximum mobility of the lower extremities in each position, the infants' clothes were removed down to the diaper and a t-shirt or 'onesie'. A 1.25 cm diameter reflective hemispherical-shaped marker was then placed on the soles of the infants' feet at the head of the first metatarsal. This marker was used during later data reduction to assist in identifying when leg movements and kicks occurred. The infants were videotaped in six positions during each data collection session. Consistent with previous research, the infants were placed



supine on a mat for the first position at each session.^{13, 15} Next, the infants were seated in a specially designed adjustable infant seat at 30, 40, 45, 50, and 60 degrees from horizontal. The infant seat was developed by Chapman (2013). (See Figure 2). Trial order for the five adjustable infant seat positions was randomized per infant for each visit. The infants were videotaped for one minute in each position. To encourage activity, the research advisor was positioned to the right of the baby to provide visual and social interaction during each trial. In addition, the parents of the infants were allowed to vocally and visually interact with their child.





Figure 2. Specially designed adjustable infant seat.

Anthropometric Data Collection

Anthropometric measurements were collected to analyze the effect that physical characteristics may have on the frequency of leg movements and kicks. Based on previous research, we measured each infant's total body length, thigh and calf lengths; body weight, circumference and skin fold measurements of the right thigh and calf, as well as right hip abduction and right ankle dorsi- and plantar-flexion.^{13, 15} We hypothesized that infants with a larger overall size would demonstrate decreased leg movements and kicks than smaller infants. In addition, infants with longer thigh and calf lengths would move their legs less often than infants with shorter legs and infants with increased joint laxity, as measured by hip and ankle range of motion, would demonstrate



decreased leg movements and kicks. Table 4 displays the technique used for collecting

the anthropometric measurements.

Measurement	Measurement Technique			
Lengths	All measurements were recorded in centimeters and measured to the nearest millimeter			
Total Body Length	The baby was supine, looking up, with the body aligned and the ankle dorsiflexed to 90 degrees. Measurement was taken from the top of the head to the bottom of the foot			
Thigh	The baby was supine. Measurements was taken from the distance betwee the greater trochanter and the lateral condyle of the femur with the leg extended and the ankle dorsiflexed to 90 degrees			
Calf	The baby was supine. The distance between the lateral condyle of the femur and the lateral malleolus with the leg extended at the hip and knee and the ankle dorsiflexed to 90 degrees			
Circumferences	All measurements were recorded in centimeters and measured to the			
Circumerences	nearest millimeter.			
Thigh	The circumference of the thigh measured at the midpoint of the segme length as measured above			
Calf	The circumference of the calf at the midpoint of the segment length as measured above			
Skinfolds	All measurements were recorded in millimeters and measured to the nearest .5mm.			
Thigh	The baby was supine. The skinfold was vertical and taken at the same level as the thigh circumference on the medial surface with the thigh extended at the hip			
Calf	The baby was supine. The skinfold was vertical and taken at the same level as the calf circumference on the medial surface with the calf extended at the knee			



Range of Motion	All measurements were recorded to the nearest degree.
Hip Abduction	The leg is abducted while the infant is supine with the knee extended. The leg is in contact with the support surface.
Ankle Plantarflexion	The infant was supine with the leg extended at the hip and knee. The ankle was moved into maximum passive plantarflexion.
Ankle Dorsiflexion	The infant was supine with the leg extended at the hip and knee. The ankle was moved into maximum passive dorsiflexion.
Weight	Measured in pounds and ounces to the nearest ounce

Data Reduction

The data was reduced using a frame by frame analysis for each trial. All researchers were trained prior to data collection and had a percent agreement rate of \geq 80% with an expert rater in identifying the frequency of leg movements and types of kicks. The researchers recorded the total number of leg movements, total number of kicks, and frequency of six types of kicks for each trial. We defined a leg movement as the moment the marker on the sole of the foot began to move until it stopped or there was a change in direction. At that point, a new leg movement began. Six specific kicks were also identified: single leg kick, single knee kick, parallel leg kick, parallel knee kick, alternating leg kick, and alternating knee kick. To be identified as a leg kick, the infant had to flex and extend their leg at the hip and knee joints. Knee kicks required there to be flexion and extension of just the knee joint. See Table 3 for a detailed description of the different forms of kicks.



Data Analysis

Due to our small sample size we chose to employ descriptive statistics to be able to review our data for trends regarding the influence that age, position, and timing of surgery had on the spontaneous kicks generated by these four babies. We began by calculating the means and standard deviations for the average number of kicks and leg movements generated in each position at each age. Then, we calculated pearson productmoment correlations between the frequency of total leg movements and kicks. We also calculated the pearson product-moment correlations between the anthropometric measures and the per minute average frequency of kicks. We ended our analysis by summarizing the average per minute frequency of kicks generated by the two infants who had their spinal lesion repaired pre-natally and the two infants who had their spinal lesion repaired shortly after they were born.

Results

A statistically significant positive relationship was found between the average frequency of leg movements and kicks generated per minute (r = .5822, $p \le .000$). These results are presented in Figure 3. Given the strength of this association, we decided to focus solely on kicks as the dependent variable when we analyzed the positional effects the specially designed adjustable infant seat had on the infants' ability to generate kicks as well as the influence of age on their ability produce kicks. Kick are especially important behaviors to study because they are kinematically similar to the leg movements used to walk.



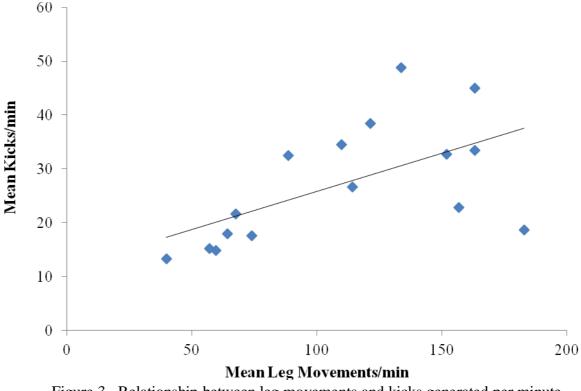
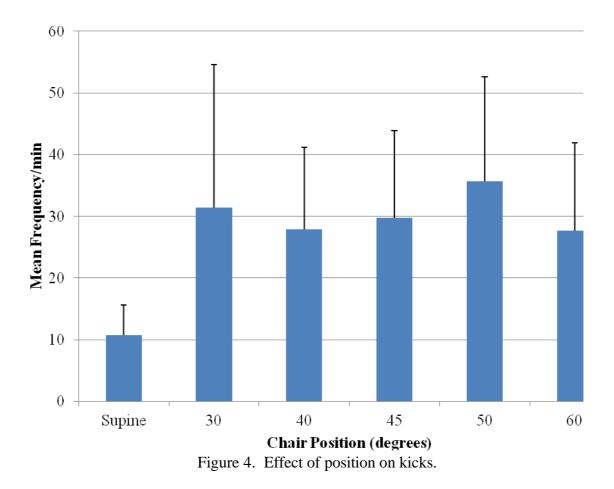


Figure 3. Relationship between leg movements and kicks generated per minute (r = 0.5822, $p \le .000$).

Due to the small sample size, the remainder of our data analysis focuses on analyzing trends in the data rather than seeking to verify the presence of significant differences. For instance, in each seated position the infants produced at least two and a half times more kicks compared to when they were supine. The means and standard deviations for kicks generated by our sample (n=4) in each position are presented in Figure 4. It is interesting to note that the infants in this study generated the greatest number of kicks per minute in the 50 degree position.





The difference in the average number of kicks generated per minute over developmental time appears to be minimal with relatively large standard deviations at each age. Figure 5 highlights these results. Thus, this data suggests that these babies with SB do not seem to increase nor decrease how often they kick their legs over developmental time.



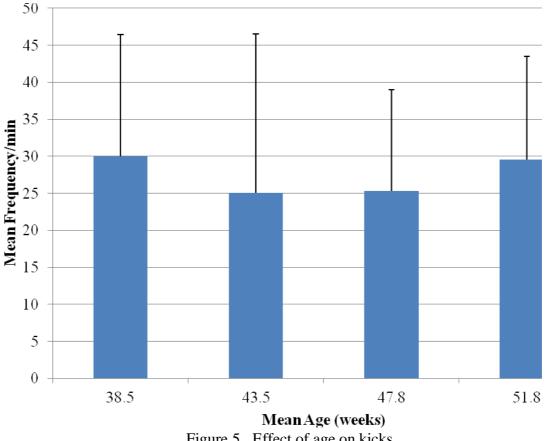


Figure 5. Effect of age on kicks.

The influence of age and position on the frequency of kicks generated per minute across developmental time by when the infant's spinal lesion was repaired are depicted in Figures 6a-f. The data presented for the pre-natal and post-natal surgery groups at the three youngest ages each have an n=2. However, at the oldest age, 51.8 weeks, only one baby in each group completed the data collection process. This is because one infant in each group began to walk between the third and fourth data collection session. As such, we were not able to collect data from those infants during their fourth visit.

When the infants were placed supine, all four infants generated nearly the same number of kicks per minute regardless of when they had their spinal lesion repaired. It is



worth noting that the two babies who had their spinal lesions repaired post-natally kicked more often at the youngest and oldest ages (See Figure 6a). These are two of the seven age and position combinations out of a possible 24 age - position combinations in which the post-natal surgery group generated more kicks per minute than the babies who had their spinal lesion repaired pre-natally.

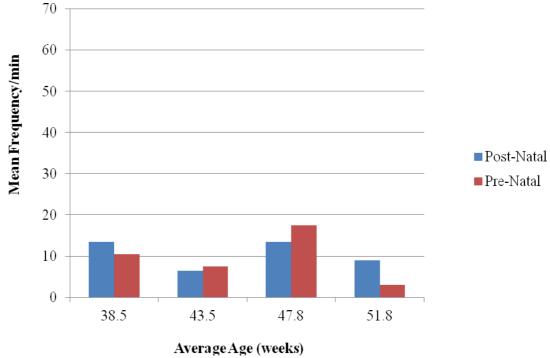


Figure 6a. Kicks generated in supine.

When the infants were placed in the 30 degree position, the infants who received the pre-natal surgery produced, on average, a greater number of kicks at each age compared to the babies who had their spinal lesion repaired after they were born. At 38.5 and 43.5 weeks of age these two infants generated markedly more kicks than when they were 47.8 and 51.8 weeks old, respectively. These results are presented in Figure 6b.



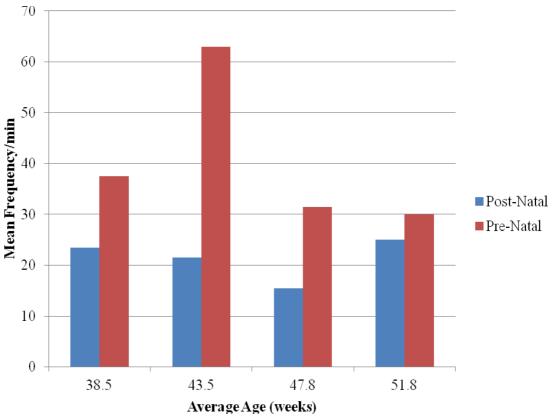


Figure 6b. Kicks generated in 30 degree position.

Figure 6c illustrates how often the infants kicked per minute when they were placed in the 40 degree position. In this position, the infants who received the pre-natal surgery kicked more often at the three youngest ages (38.5, 43.5 and 47.8 weeks of age) compared to the babies who had their spinal lesion repaired post-natally. This trend was reversed at the oldest age (51.8 weeks of age).



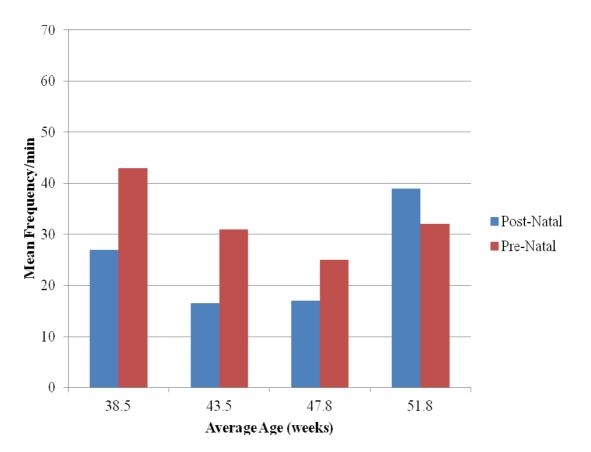


Figure 6c. Kicks generated in 40 degree position.

When the infants were placed in the 45 degree position, the infants who had their spinal lesions repaired pre-natally generated, on average, a greater number of kicks at the two youngest ages compared to the babies who had their spinal lesion repaired after birth. This trend was reversed when the infants were 47.8 and 51.8 weeks old, respectively. At these two ages, the babies who had their spinal lesion repaired post-natally generated more kicks than the babies who had their spinal lesion repaired in-utero. Figure 6d highlights these results.



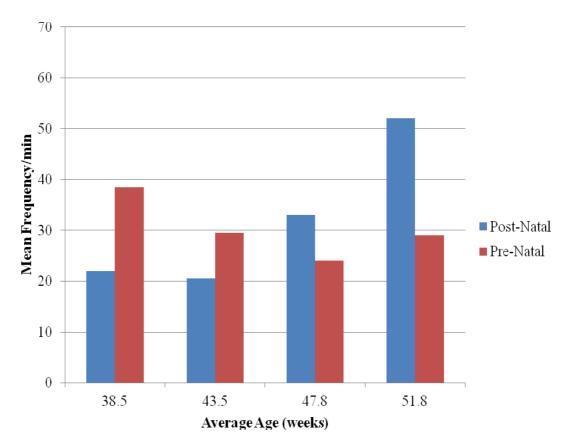


Figure 6d. Kicks generated in 45 degree position.

When the infants were placed in the 50 degree position, the infants who underwent the pre-natal surgery produced almost the same number of kicks per minute when they were 38.7 weeks old compared to the babies who experienced the post-natal surgery. On average, they generated a greater number of kicks at the two middle ages (43.5 and 47.8 weeks) compared to the infants who underwent the post-natal repair. At 51.8 weeks of age, the baby who had her spinal lesion repaired after birth generated more kicks than the baby who underwent the pre-natal repair procedure. This results are depicted in Figure 6e.



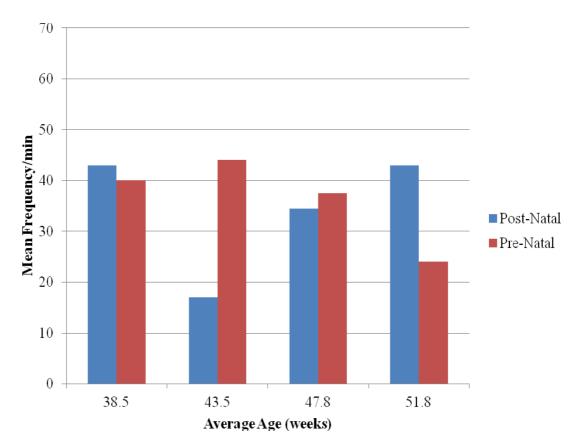


Figure 6e. Kicks generated in 50 degree position.

Figure 8f shows our observations that when the infants were placed in the 60 degree position those who had their spinal lesion repaired pre-natally produced more kicks on average per minute compared to those who had their spinal lesion repaired post-natally at all four ages.



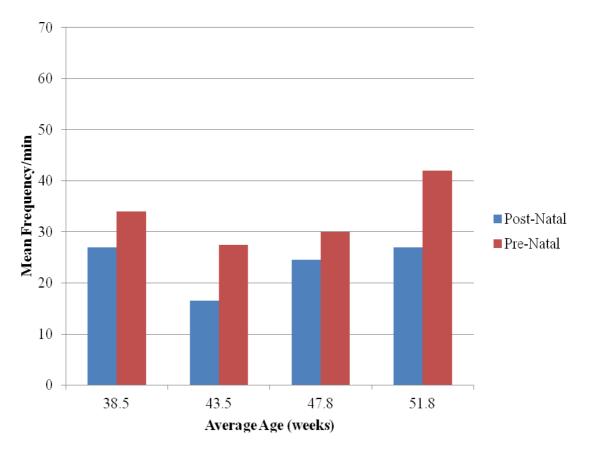


Figure 6f. Kicks generated in 60 degree position.

Table 5 presents the chronological age each infant achieved a given developmental milestone. There does not seem to be a clear trend when babies achieve their developmental milestones based on when they had their surgery.



Participant	Roll over	Sit	Crawl	Pull to stand	Cruise	Walk
1	4 months		7.5 months	8 months	11 months	12 months
2	8 months	12 month				
3	9.75 months	8.5 month	11 months	12.5 months	12.75 months	13 months with walker
4	4 months	7 months		7.5 months at furniture		

Table 5. Developmental Milestones.

Table 6 presents the relationship between the anthropometric data we collected and how often the babies in this study produced kicks. We found significant correlations between plantarflexion and kicks as well as thigh skinfold and kicks. No other correlations reach significance, but dorsiflexion approached significance, (p=.062).



Anthropometric Measure	R value with Kicks	Significance Level
Height	-0.017	0.949
Thigh Length	-0.161	0.550
Calf Length	0.193	0.473
Thigh Circumference	0.215	0.424
Thigh Skinfold	0.636	0.008
Calf Circumference	-0.003	0.990
Calf Skinfold	0.227	0.398
Weight	0.209	0.437
Dorsiflexion	-0.476	0.062
Plantarflexion	0.667	0.005
Hip Abduction	0.328	0.215

Table 6. Relationship between Anthropometric Data and Kicks.

Discussion

Our results suggest that all four infants with SB appear to be sensitive to changes in their position as they altered how often they kicked and how often they generated certain kinds of kicks, e.g. parallel knee kicks as they confronted changes in their movement context. The infants in this study generated more kicks in all six positions of the specially designed adjustable infant seat compared to when they were supine. This is



consistent with Chapman's research in which he reported that infants with SB generated significantly more leg movements and kicks when they were seated in a specially designed infant seat set at 45 degrees from horizontal compared to when they were supine or seated in a conventional infant seat.^{13, 15} Increasing the number of kicks generated per minute strengthens the neural connections linked with the sensory information associated with kicking as well as the muscles used to kick. We may be able to enhance the motor development process of infants with SB by placing them in positions that will allow them to reinforce the efferent and afferent neural connections associated with kicks generated during the first year of life and prior to when these babies begin to walk.

These data also support the principle of self-organization. The observations that these four infants with SB increased and/or decreased how often they produced kicks depending on the position they were placed in and altered how often they generated particular types of kicks, e.g. parallel knee kicks offers additional support for the idea that babies with SB are capable of self-organizing their multiple sub-systems as they confront changes in their movement context. ^{13, 15}

How often infants with SB produce leg movements and kicks may be considered a rate-limiter or control parameter depending on how we use the movement context to influence how often babies with SB move their legs and kick. If we place them in positions that decrease how often they move their legs and kick then, we are, in effect creating a rate-limiting situation that prevents them from strengthening the leg muscles needed to kick as well as weakening the neural connections that support kicks. Alternatively, if we place babies with SB in positions that enhance their ability to move



their legs and kick we have created a control parameter for them. In essence, our data shows that we can use the position we place babies in to facilitate their ability to kick more often and generate more kicks of a certain type, e.g. single knee kicks. Research with other groups of babies, e.g. babies with Down syndrome and typically developing infants suggests that how often babies kick is correlated with then babies begin to walk.²²

We did not observe a position(s) that led to an increase or decrease in kicks over developmental time. These babies, like other groups of infants with lumbar SB, seem to have their own self-selected rate of kicking that does not change over developmental time.^{13, 15} This singular observation is consistent with the existing literature and is important because parents, therapists and other providers need to be aware that most babies with lumbar SB do not follow the same developmental path for leg movements and kicks that TD babies do prior to when they begin to walk. Our data coupled with the current literature suggests that most infants with lumbar SB, regardless of the movement context in which they are placed, do not increase nor do they decrease how often they move their legs and kick over developmental time like babies who are developing typically.

We specifically addressed the timing of the surgical procedure to repair the spinal lesion because we were fortunate enough to recruit two babies who had their spinal lesion repaired pre-natally. This is extremely important because there is a marked lack of data about how infants with SB who have their spinal lesion repaired pre-natally develop motor skills. The two infants who had in-utero surgery demonstrated more kicks in each seated position at each age compared to the infants who had post-natal spinal surgery.



Both the prenatal and postnatal group generated similar numbers of kicks when they were supine, but the pair who had their spinal lesion repaired pre-natally generated 35% more kicks compared to the two babies who had their spinal lesion repaired post-natally in each seated position. It appears that infants with SB who have their spinal lesion repaired pre-natally have an increased sensitivity to changes in their position which enabled them to kick more often at each age that babies who had their spinal lesion repaired post-natally. This type of increased sensitivity may positively influence the quantity and/or quality of the sensory feedback available to these infants, which may contribute to the neural pathways for kicks being strengthened. This phenomenon may lead to these babies walking earlier in life compared to babies who have their spinal lesion repaired after birth. However, there is limited empirical data at this point in time to support this claim.

One additional factor that we considered is the influence that body characteristics, like leg length or girth may have on how often infants with SB kick. The anthropometric data, similar to Chapman's earlier work, demonstrated low to moderate correlations for all measurements except thigh skinfold with kicks as well as plantarflexion with kicks.^{13,} ¹⁵ These measurements were significantly correlated with leg movements and kicks; however, due to our small sample size we are unsure of how these characteristics contribute to how often infants with SB kick. Our hypothesis was that the smaller the infants' legs the easier it would be for them to kick but our current research suggests that larger thigh skinfold measurements are related to how often infants with SB kick. Additionally, we observed that increased plantarflexion was associated with more leg movements and kicks. Further research and recruitment of more subjects may lead to a



better understanding of the relationship between selected anthropometric traits and how often babies with SB kick.

Limitations

The small sample size limited our power in performing a formal statistical analysis. Further recruitment will be necessary to complete a formal MANOVA with repeated measures analysis to verify any significant differences for position, age and timing of the surgical repair. This may also help determine if there is a correlation between the frequency of kicks and age of independent ambulation as well as other possible benefits for motor development.

Additionally, the age range of 8 months to 15 months only gives us a small window of time to observe changes in leg movements and kicks over time. Infants with SB at different ages may show different results than our current data. Additional research on the association between kicks and motor development may give us a better idea if the frequency of kicks correlates earlier ambulation.

Conclusion

In light of our current results and previous literature pertaining to infants with SB between 4 and 15 months of age, we believe that position can be used to facilitate the frequency of kicks in infants with SB. Any seated position in the specially designed adjustable infant seat led to an increase in kicks when compared to supine. Additionally, infants with lumbar SB seem to have their own preferred rate of kicking that does not change over developmental time. This is different from typically developing babies who increase their rate of kicking until they pull to stand at which time they decrease how



often they kick in all positions. Finally, the infants who had their spinal lesions repaired prenatally appear to be more sensitive to changes in position, as evidence by the increased number of kicks they generated compared to the babies who did not have this surgery before they were born. This suggests that repairing the spine prenatally may have a positive impact on motor development during the first year of life. However, further research needs to be completed to determine if there is a strong, positive correlation between the frequency of kicks and age of independent ambulation.



References

1.Centers for Disease Control. *Spina Bifida.* http://www.cdc.gov/ncbddd/spinabifida/data.html. Accessed 10/26/13.

2.Campbell SK, Vander Linden DW, Palisano RJ. (2011). *Physical Therapy for Children*, 4th Ed. Saunders Elsevier: Philadelphia, PA.

3. Chapman DD. Context Effects on the Intrinsic Dynamics of Infants with Spina Bifida (SB). Bloomington: Indiana University; 1997.

4. Hobbins, J.C., Gannuan, P.A.T., Berkowitz, R.L. (1979). Ultrasound in the diagnosis of congenital anomalies. *American Journal of Obstetrics and Gynecology*, 134, 131.

5.Spina Bifida Association, FAQ about Spina Bifida. http://www.spinabifidaassociation.org/site/c.liKWL7PLLrF/b.2642327/k.5899/FAQ _About_Spina_Bifida.htm. Updated 2011. Accessed 10/26/13.

6. Children's Hospital of Philadelphia. Spina Bifida. http://www.chop.edu/service/fetal-diagnosis-and-treatment/fetal-diagnoses/spina-bifida.html. Accessed 11/1/13.

7. Berger, S.E., Theuring C., Adolph, K.E. How and when infants learn to climb stairs. *Infant Behav Dev Infant Behav Dev.* 2007;30(1).

8. McGraw, MB. Neuromuscular development of the human infant as exemplified in the achievement of erect locomotion. J. of Ped., 17:747-771, 1940.

9. McGraw, MB. *The Neuromuscular Maturation of the Human Infant*. New York: Hafner Press, 1940.

10. Gulsvig, Katie; Hawn, Christina; Plummer, James; and Schmitz, Ann, "The Sensitivity of Infants with Spina Bifida to Sensory Information" (2012).Doctor of Physical Therapy Research Papers.

11. Thelen E, Ulrich BD. Hidden Skills: A Dynamic Systems Analysis of Treadmill Stepping During the First Year. *Monogr Soc Res Child*. 1991;56(1).

12. Thelen E. Treadmill-elicited stepping in seven-month-old infants. *Child Dev.* 1986;57:1498-1506.

13. Chapman D. Context effects on the spontaneous leg movements of infants with spina bifida. *Pediatr Phys Ther.* June 2002;14(2):62-73.



14. Thelen E., Fisher DM., Ridley-Johnson R. (1984). The relationship between physical growth and a newborn reflex. *Inf Beh & Dev.* 7, 479-493.

15. Chapman D. Context effects on the ability of young infants with myelomeningeocele to generate complex pattern leg movements. *Pediatr Phys Ther.* Summer 2009;14(2):62-73.

16. Edelman, GM. *Neural Darwinism: The Theory of Neuronal Group Selection*. New York: Basic Books, Inc. Publishers, 1987.

17. Edelman, GM. *The Remembered Present*. New York: Basic Books, Inc. Publishers, 1989.

18. Edelman, GM & Mountcastle, VB. *The Mindful Brain*. Cambridge, MA: The MIT Press. 1978.

19.Ulrich BD, Ulrich DA. Spontaneous Leg Movements of Infants with Down Syndrome and Nondisabled Infants. *Child Dev.* 1995;66(6):1844-1855.

20. Sival, D.A., Brouwer, O.F., Bruggink, J.L., Vles, J.S., Stall-Schrieninemachers, A.L., Soller., K.M. ... Bos, A.F. (2005). Movement analysis in neonates with spina bifida aperta. *Early Human Development*, 82(4), 227-234.

21. Rademacher, N., Black, D.P., & Ulrich, B.D., (2008). Early spontaneous leg movements in infants born with and without myelomeningocele. *Pediatric Physical Therapy*, 20(2), 137-145.

