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Goal-directed Leg Movements and Kicks in Infants with Spina Bifida

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ABSTRACT

BACKGROUND AND PURPOSE: Infants with SB present with a known central nervous system lesion that often results in neurologic, orthopedic, and/or cognitive impairments. They usually learn to walk significantly later than typically developing (TD) infants. The delays they experience in learning to walk appear to be related to the fact that they move their legs and kick less often than infants who are TD. Only a small number of studies have reported strategies that therapists and parents may use to increase how often infants with SB move their legs and kick. The purpose of this study was to examine the impact conjugate reinforcement has on the frequency of leg movements (LMs) and kicks generated by infants with SB.

METHODS: The LMs of 7 infants with lumbar or sacral SB were videotaped while they were supine in 3 conditions: Baseline; Acquisition (tethered to a mobile); and Extinction. The infants' LMs were video-taped for two minutes in each condition which enabled us to capture their spontaneous and goal-directed LMs and kicks. The video-tape of each infant's LMs were then behavior coded via a frame by frame analysis to identify how often each baby moved his or her legs and kicked in each condition as well as how often they generated 9 types of kicks.

RESULTS: A significant correlation was observed between LMs and kicks ($r= .976$, $p=0.00$). These infants moved their tethered leg significantly more than their untethered leg ($p=0.036$). These infants generated more goal-directed LMs and kicks in the acquisition and extinction conditions; however, these differences only approached significance ($p \leq .05$). Single kicks and parallel kicks were the most common types of kicks generated in each condition.

CONCLUSION: The present results are consistent with the literature and suggest that increased kicks lead to stronger neural connections and increased strength, which ultimately leads to earlier onset of ambulation. Due to the significant correlation between LMs and kicks, increasing the frequency of LMs in infants may increase the amount of kicks. However, further research is needed. This study was limited by the small sample size and large standard deviations within group means.

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

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

Primary Advisor David Chapman Date 4/26/15.

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Introduction

Spina bifida (SB) is the most common neural tube birth defect.[1,2] In the United States, approximately 1,400-1,500 infants are born with SB each year, which translates to approximately eight infants per day.[1,3] This includes the 30-40 babies born with SB each year in Minnesota.[4] Spina bifida means "split spine" and occurs when the posterior halves of the vertebral arch fail to fuse during the fourth week of embryogenesis.[1,5] As a result, a sac of spinal tissues may protrude dorsally through this opening. This puts these tissues, e.g. nerve roots, at risk for damage. Lesions may occur at any level of the spine, but most commonly occur in the lumbar and sacral regions.[6]

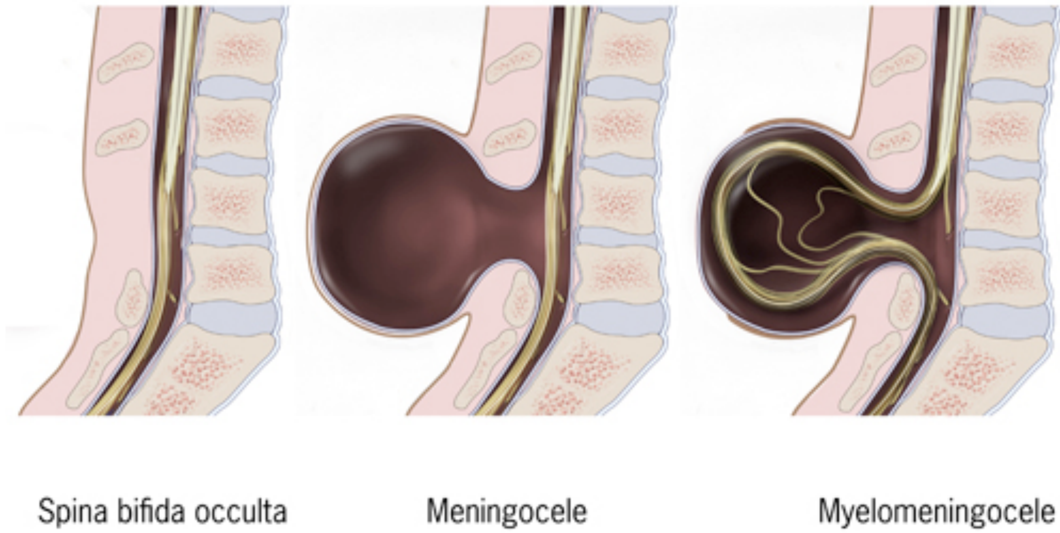
The specific cause of SB is unknown, but is theorized to be the result of a combination of genetic and environmental factors.[1] In addition, several risk factors have been identified, which increase the probability of a child being born with SB.[7] A lack of folic acid intake during pregnancy has been correlated with an increased risk of SB.[2] As a result, the United States Public Health Service's and the Centers for Disease Control recommend that women of childbearing years, generally between the ages of 15-45, consume 400 micrograms of folic acid each day as a preventative measure. Due to the fact that the development of SB occurs between the third and fourth week of pregnancy, it is important to not wait until a woman knows she is pregnant to begin taking folic acid. Otherwise, it may be too late to prevent a spinal lesion from developing.[8] Other risk factors for SB include the consumption of anticonvulsant medications during pregnancy, being of Caucasian or Hispanic race, a female embryo/fetus, having a family history of SB, uncontrolled diabetes, maternal obesity and maternal hyperthermia.[9,10]

There are three distinct types of SB, which range in severity and are illustrated in Figure 1. The mildest form is spina bifida occulta (SBO), which is referred to as 'hidden spina bifida' because as many as 15% of healthy individuals have it, but may be unaware.[1] Usually an individual with SBO presents with no impairments or spinal cord abnormalities. A small portion of these individuals may, however, demonstrate pain or other neurological symptoms. A rare few experience a tethered spinal cord, which may cause bowel and/or bladder dysfunction.

The other two types of SB are meningocele and myelomeningocele. Together, these forms of SB are classified as SB aperta. Meningocele is less severe than myelomeningocele. In meningocele, part of the meninges of the spinal cord push through the spinal opening and present as a sac-like structure on the dorsal side of the body. Although the opening is large enough for the meninges to protrude, the sac contains only cerebrospinal fluid. This results in little to no damage to the spinal cord or nerve roots.[7] Individuals with this type of SB may present with impairments ranging from minimal to those seen in the more severe myelomeningocele form of SB.

The most severe form of SB is myelomeningocele. Myelomeningocele occurs when the spinal cord and nerves protrude posteriorly through the portion of the spinal vertebrae that failed to close. In most cases, this causes nerve damage, which ultimately may lead to impairments and disability. Approximately 70-90% of infants born with this form of SB also experience hydrocephalus, which is an accumulation of excess cerebrospinal fluid in the brain.[1]

Figure 1. The Three Types of Spina Bifida[8]



Infants born with myelomeningocele often experience sensory, motor and cognitive impairments.[7,11] In the field of physical therapy, we primarily focus on the musculoskeletal impairments that a child presents with, such as clubfoot, hip dysplasia, increased risk of osteoporosis, and motor paralysis. Other impairments include those with a sensory component, including a neurogenic bowel and bladder, cranial nerve palsies, and visual-perceptual deficits. Cognitive deficits and learning disabilities may also become evident as the infant develops. As a result of these impairments, which are displayed in Table 1, infants with SB experience significant delays in their motor development.[10] In general, it has been found that infants with higher spinal lesions demonstrate greater and more severe impairments than infants with lower lesions.

Table 1. Common Impairments Experienced by Infants with SB

Motor	**Sensory**	**Cognitive**
Paralysis	Neurogenic bowel and bladder	Language
Increased risk of osteoporosis	Cranial nerve palsies	
Spasticity	Visual-perceptual deficits	
Musculoskeletal deformities	Latex allergy	
Upper limb discoordination	Skin breakdown	

The medical management of SB begins during prenatal development.[1] It may first be detected via an alpha-fetoprotein blood test that is usually performed between 16-18 weeks of gestation. The sensitivity of this test is moderate to high, as the protein is higher in about 75-90% of women who are carrying a fetus with SB.[12] Spina bifida is also commonly detected during routine ultrasound examinations. Finally, it may be confirmed during a maternal amniocentesis during which amniotic fluid is extracted from the womb through a needle and tested for abnormalities of protein levels. When a fetus is diagnosed with SB a decision needs to be made by the parents about whether they will have the spinal lesion surgically repaired prenatally or after the baby is born.

In-utero surgery is an innovative procedure, which was first performed in 1998. In-utero surgical repair has been found to result in decreased incidence of hydrocephalus and the need for shunt placement as well as improvements in overall mobility and likelihood that a child will ambulate.[3] The decision to undergo in-utero surgical intervention is not an easy one as it is currently only available at specific locations across the country, and the mother must remain on bed rest for the remainder of her pregnancy. If the surgery is performed post-delivery, it usually occurs within 24-48 hours of birth. Regardless of the timing of surgery, a cesarean section is also scheduled to reduce the trauma to the infant during delivery.

After birth, an infant may experience a variety of impairments as mentioned above and summarized in Table 1. In addition, infants with SB often experience hydrocephalus, which is typically treated with the placement of a ventriculo-peritoneal (VP) shunt.[8] The shunt is a small hollow tube that helps drain excess cerebrospinal fluid from the baby's brain and into the abdominal cavity to reduce intracranial pressure. If hydrocephalus goes untreated, it can cause injury to the brain itself. Musculoskeletal impairments, i.e. clubfoot, can be managed with serial casting of the involved foot/feet and/or ankle-foot orthoses (AFOs). Serial casting entails a series of casts that are applied and removed weekly, which slowly reduces the musculoskeletal deformation of the foot/feet. Ankle-foot orthoses may be prescribed independent of and/or following serial cast treatment and are usually worn until the foot/feet are in a stable position. The treatment goal is to improve the alignment of the foot and ankle, which typically enhances the child's ability to stand and walk. Delays in motor development are usually treated through physical therapy intervention.

Along with experiencing the impairments noted above, infants with SB often achieve motor milestones later than typically developing (TD) infants.[6,7,10] As shown in Table 2, all major motor milestones, from sitting and standing to crawling and walking, are delayed. For example, TD infants usually begin to walk when they are between 12-15 months old, while infants with SB learn to walk some time between 3 to 7 years, on average.[7]

Table 2. Developmental Milestones

Developmental Milestone	Typically Developing Infant	Infants with SB
Sitting	6-7 months	1-2 years
Crawling	7-11 months	1-2 years
Standing	9-13 months	3 years
Walking	12-15 months	3-7 years

Collectively, the literature cited above suggests that infants with SB may experience multiple types of impairments and that they often demonstrate delays in their motor development.[6,7,10,13] However, what is less clear from the literature reviewed above is an explanation of why infants with SB usually acquire new motor skills later in life than TD babies.

Historically, therapists and developmentalists have suggested the delays infants with SB experience in their motor development are related to the level of their spinal lesion, i.e. infants with higher spinal lesions develop new motor skills later in life than infants with lesions that are located lower in the spine.[6,10] This school of thought follows the neuromaturation approach to motor development which postulates that new motor skills are acquired as a result of maturation of the central nervous system (CNS) and in a specific order, e.g. babies crawl before they walk as well as from head to toe and proximal to distal.[14,15]

More recent research, guided by the concept of neuroplasticity and the principle of self-organization from the dynamic systems theory has shown that the neuromaturation approach does not account for individual differences across infants nor does it help parents and therapists understand how other factors, e.g. the position in which infants are placed, their movement experiences or feedback from the environment, may influence when infants with SB learn new motor skills.[6,10,13,16,17,18,19]

Neuroplasticity describes the ability of neurons to change their chemical profile, structure or function in response to stimuli.[20] Proponents of neuroplasticity suggest that the neural connections that support a given behavior, e.g. reaching with one arm or kicking with the legs, are strengthened in real time as an individual reaches with her preferred arm or repeatedly kicks her legs.[21,22] Ultimately, stronger neural connections are thought to increase the likelihood that a given behavior will be demonstrated in the future. For example, an infant who repeatedly kicks with 1 leg rather than 2 would be more likely to kick with 1 leg in the future. Alternatively, a baby who frequently demonstrates alternating kicks would be more likely to generate alternating kicks in the future compared to the child who kicks with only one leg.

Thelen and her colleagues at Indiana University were among the first to apply the principle of self-organization to the study of infant motor development.[10,17,18,23] Self-organization means that new motor skills emerge from the interactions of multiple body systems within a given environment and context rather than by hard-wired neural templates. Advocates of self-organization believe that internal, i.e. intrinsic body systems like muscles and external factors, and a child's environment work together to influence how and when new motor skills develop. For example, for a child to successfully reach for a given object she needs sufficient strength, upper extremity range of motion, vision and motivation/cognition as well as an attractive toy to be placed within her environment. Thus, supporters of this approach suggest that infants organize their multiple subsystems to create a movement that meets the demands of a task within a given context.

Thelen illustrated the principle of self-organization in a simple, yet profound study in which she observed how infants, who no longer demonstrated the infant stepping response, stepped when they were held chest deep in water.[24] She found that infants who did not demonstrate the infant stepping response on a firm surface were able to demonstrate alternating steps when they were held in the buoyant water environment. These results showed for the first time that the environment can significantly influence an infant's ability to move, and implied that the nervous system is not the only factor that influences how babies move their arms and legs.

More recently, researchers have utilized the concept of neuroplasticity and the principle of self-organization to examine how infants with SB learn to coordinate their leg movements during their first year of life. For instance, controlled observations of infants with SB show that during their first week of life infants with SB reduce how often they move their legs when they are placed in a supine position.[25] Systematic observations of the spontaneous leg movements of infants with SB at 1, 3, 6, and 9 months show that they demonstrate fewer leg movements that are shorter in duration and less complex when supine compared to age-matched TD infants.[26,27] Four to 7 month old infants with lumbar or sacral lesions also spontaneously move their legs and kick less often than age-matched TD infants when they are placed supine, in a specially designed infant seat, and in a conventional infant seat.[6,10,13] These infants did not increase nor did they decrease how often they moved their legs and kicked between 4 and 7 months of age. But, they did move their legs and kick significantly more often when they were seated in the specially designed infant seat compared to when they were supine or seated in the conventional infant seat. Older infants with lumbar or sacral SB between 7 and 11 months of age also spontaneously moved their legs more often when they were seated in the specially designed infant seat compared to when they were placed in a conventional infant seat.[19] In addition, they generated significantly more kicks when seated in the specially designed infant seat compared to when they were placed supine or positioned in a conventional infant seat.

These studies have focused on how often infants with SB generate spontaneous leg movements and kicks during their first year of life. Spontaneous leg movements occur when an infant moves her legs in the absence of a particular or observable goal. This set of studies has also centered on the effect the infants' position has on their ability to move their legs and kick. Clearly, they show that infants with lumbar or sacral SB are sensitive to their position in space and that an infant's position may be used to promote or hinder her ability to spontaneously move her legs and kick.[6,10,13,19] They also suggest that infants with SB may need direct interventions if they are going to increase how often they move their legs and kick over developmental time. These studies do not, however, reveal if infants with SB are capable of generating goal-directed leg movements and kicks. Are they sensitive to visual and/or auditory feedback that is directly linked to their leg movements and kicks? Will infants with SB increase how often they move their legs and kick if they are provided visual and/or auditory feedback that is directly related to their leg movements and kicks?

Goal-directed leg movements and kicks, in contrast to spontaneous leg movements, are leg movements that achieve a goal or result in a desired outcome. For example, Rovee and Rovee and Collier reported that young infants with and without Down syndrome (Ds) generated more leg movements when they were tethered to an overhead mobile than they did in a baseline condition in which they spontaneously moved their legs and kicked.[28,29] This paradigm, known as conjugate reinforcement, can be implemented when infants are provided with the opportunity to achieve a desirable outcome, e.g. make an overhead mobile move via a particular behavior, such as leg movements or kicks. To date, only one small pilot study (n=3) has been completed that examined the impact of conjugate reinforcement on the leg movements and kicks of infants with SB. In this pilot project, Chapman and colleagues found that being tethered to an overhead mobile had a positive effect on the frequency of leg movements and kicks generated by infants with SB.[30] But, they could not generalize their findings due to their small sample size. Thus, we do not know if infants with SB will generate more goal-directed leg movements and kicks than they do spontaneously. The primary focus of this study was to compare how often infants with SB produce goal-directed leg movements and kicks versus how often they spontaneously move their legs and kick.

Methods

Participants

The infants in the study were recruited through the SB clinic at Gillette Children's Hospital in Saint Paul, Minnesota following Institutional Review Board approval. Eight infants participated in data collection with seven infants successfully completing the study. The data from one infant was not useable because the infant repeatedly grabbed the ribbon during the acquisition trial. Four of the infants were female and three were male. Their ages ranged between 16 and 42 weeks and 5 had ventricular-peritoneal (VP) shunts. Several of the infants presented with one or more musculoskeletal impairments commonly found in infants with lumbar or sacral SB. None of the infants presented with a known auditory or visual impairment. Table 3 displays the characteristics of each participant. Each subject and his or her parents received a participation incentive in the form of a $10.00 Target gift card.

Table 3. Infant Characteristics

Infant	Age when Tested (wks)	Gender	Lesion Level	Current Medication	Surgical History	Orthopedic Impairments	Shunt Placed
1	16	M	L4-5[a]	None	R Achilles release	Right clubfoot	VP
2	36	M	L4	None	Bilateral Achilles release	Bilateral clubfoot	VP
3	32	F	L4-5	None	None	Right hip dislocated, Left hip subluxed	VP
4	28	F	L3-5	None	Bilateral Achilles release	Atypical pelvis, Bilateral hip dislocated, Bilateral clubfoot	VP
5	24	F	S1-2	None	None	None	No
6	36	F	L5-S4	None	Right Achilles release	Right clubfoot, Right hip subluxed	No
7	42	M	L4	None	Bilateral Achilles release	Bilateral clubfoot	VP

Movement Data Collection

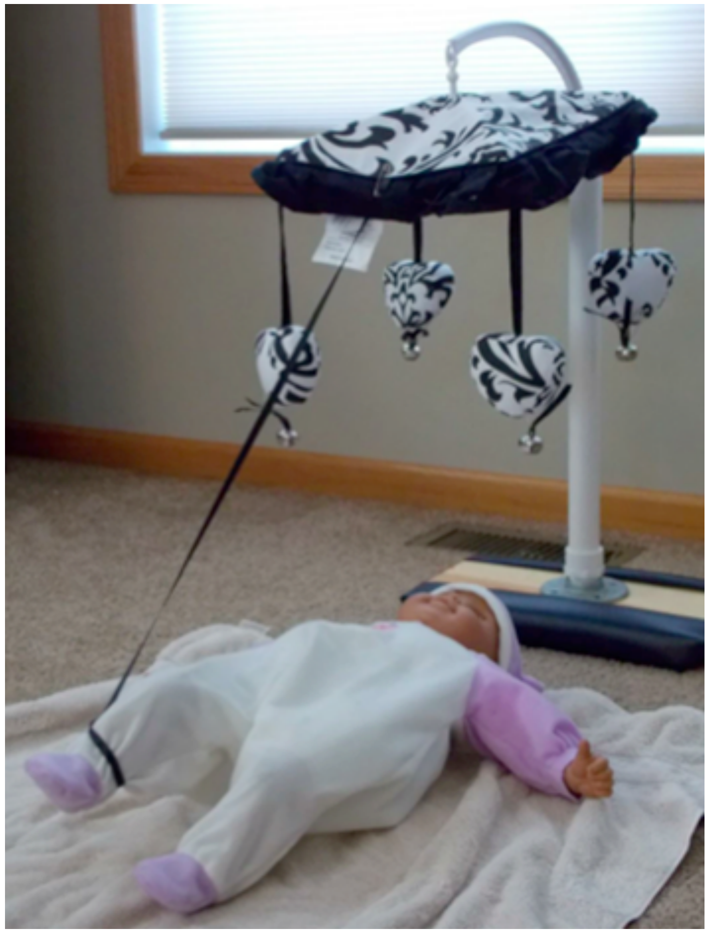
Data were collected in each infant's home with the parents present when the infant was awake, alert, and active. Based on previous research two-dimensional data was collected with a Sony handycam at 30 Hz, because the goal of this study was to record the number of leg movements and kicks rather than the kinetic properties of the infants' leg movements and kicks.[6,]

[7,10,19] The camera was placed approximately 1.5 meters from and perpendicular to the space the infant would occupy during each trial. This resulted in a camera angle that was approximately 23 degrees above the floor. Prior to videotaping the babies' leg movements, a one-meter calibration stick was placed on a small wooden platform parallel to the floor and videotaped for one minute.

During data collection, the infant wore a diaper and a t-shirt or 'onesy' to allow for increased freedom of movement. A small circular marker, approximately two centimeters in diameter, was placed on the head of the first metatarsal on the plantar aspect of each foot. The infant was then placed in supine on a towel under an overhead mobile that had four jingle bells attached to it. The mobile was positioned at a height that prevented most of the infants from reaching the mobile with his or her arm and hand.

Each infant's leg movement data were collected during three different conditions: baseline (BL), acquisition (ACQ), and extinction (EXT). Each condition was two minutes in length. A silk ribbon was tethered to one of the infant's ankle during all three conditions. During BL, the ribbon was not connected to the mobile. As such, the mobile did not move when the infant moved his or her legs and/or kicked in BL. In the ACQ condition, the ribbon was tied to the infant's ankle and the overhead mobile. When the infant moved or kicked his or her leg(s), the mobile moved and provided visual and auditory feedback to the baby.[28,29] During the EXT condition, the ribbon was tethered only to the baby's ankle, like in the BL condition. Between each condition, the infant was given a short rest break. The choice of which leg was tethered to the mobile was randomly selected for each baby. While the data was being collected, the parents were seated near their child so that they did not obstruct the infant's line of vision to the mobile. Figure 2 depicts a mockup of the data collection design.

Figure 2. Data collection set-up



Based on previous research, all kicks and leg movements during BL were conceptualized as spontaneous because when the infant moved his or her leg, the mobile did not move in response.[7,10] The ACQ condition resulted in goal-directed leg movements and kicks because the infant was tethered to the overhead mobile and could potentially learn that his or her leg movements and kicks resulted in visual and auditory stimuli. The EXT condition also resulted in goal-directed leg movements and kicks because the infant had previously experienced how his or her leg movements and kicks influenced the mobile during the ACQ condition.

Data Reduction

The total number of leg movements and kicks for each of the three conditions were behavior coded via a frame-by-frame analysis using the videotaped record of each baby's leg movements and kicks. Before behavior coding data, the student researchers achieved an 85% agreement with an expert rater for identifying kicks. The student researchers were responsible for behavior coding kicks. The research advisor behavior coded leg movements. The definition of a leg movement was when the marker on the bottom of the infant's foot started, stopped or changed directions.[6,7,10,13,19,31] There were three main categories of kicks that were coded: single, parallel or alternating.

Single kicks consisted single leg kicks, single hip kicks, or single knee kicks.[7,10,19] Single leg kicks occurred when there was flexion and extension of one leg at the hip and knee joints. Single hip kicks occurred when there was flexion and extension of one leg at the hip joint. Single knee kicks were observed when there was flexion and extension of one leg at the knee joint.

Parallel kicks occurred in a bilateral fashion where the legs moved in unison.[7,10] As with single kicks, parallel kicks consisted of parallel leg kicks, parallel hip kicks, and parallel knee kicks. Parallel leg kicks occurred when there was flexion and extension of both legs at the hip and knee joints. Parallel hip kicks occurred when there was flexion and extension of both legs at the hip joints. Parallel knee kicks occurred when there was flexion and extension of the knee joints of both legs.

Alternating kicks are the last type of kicks to be coded and occurred when both legs moved in a reciprocal pattern.[7,10] Alternating leg kicks were observed when there was reciprocal flexion and extension at the hip and knee joints of both legs. Alternating hip kicks occurred when there was reciprocal flexion and extension of the legs at the hip joints. Finally, alternating knee kicks occurred when there was reciprocal flexion and extension of the legs at the knee joints. Table 4 describes each type of kick that was behavior coded.

Table 4. Description of Kicks

Categories of Kicks	Specific Kick Types	Unilateral or Bilateral	Joints Involved	Motions Occurring
Single Kicks	Hip	Unilateral	Hip	Flexion & Extension
	Knee		Knee	
	Leg		Hip & Knee	
Parallel Kicks (in unison)	Hip	Bilateral	Hip	Flexion & Extension
	Knee		Knee	
	Leg		Hip & Knee	
Alternating Kicks (reciprocal)	Hip	Bilateral	Hip	Flexion & Extension
	Knee		Knee	
	Leg		Hip & Knee	

Data Analysis

The data analysis began by calculating a Pearson R correlation between the frequency of leg movements and kicks. A paired t-test was then used to compare the frequency of leg movements of the tethered leg to the untethered leg in the acquisition condition. A paired t-test was also used to compare the frequency of spontaneous and goal-directed leg movements and kicks in the BL versus the ACQ or EXT condition with the highest number of leg movements and kicks. All statistical analyses were completed with SPSS Version 22.

Results

Our data analysis began by graphing the raw data for leg movements and kicks and then calculating the Pearson R correlation between these two dependent variables. The significant correlation observed between the frequency of leg movements and kicks $\{r = 0.976, p = 0.00\}$ is displayed in Figure 3. Within this group of infants with lumbar or sacral SB, infants who produced more leg movements also produced more kicks.

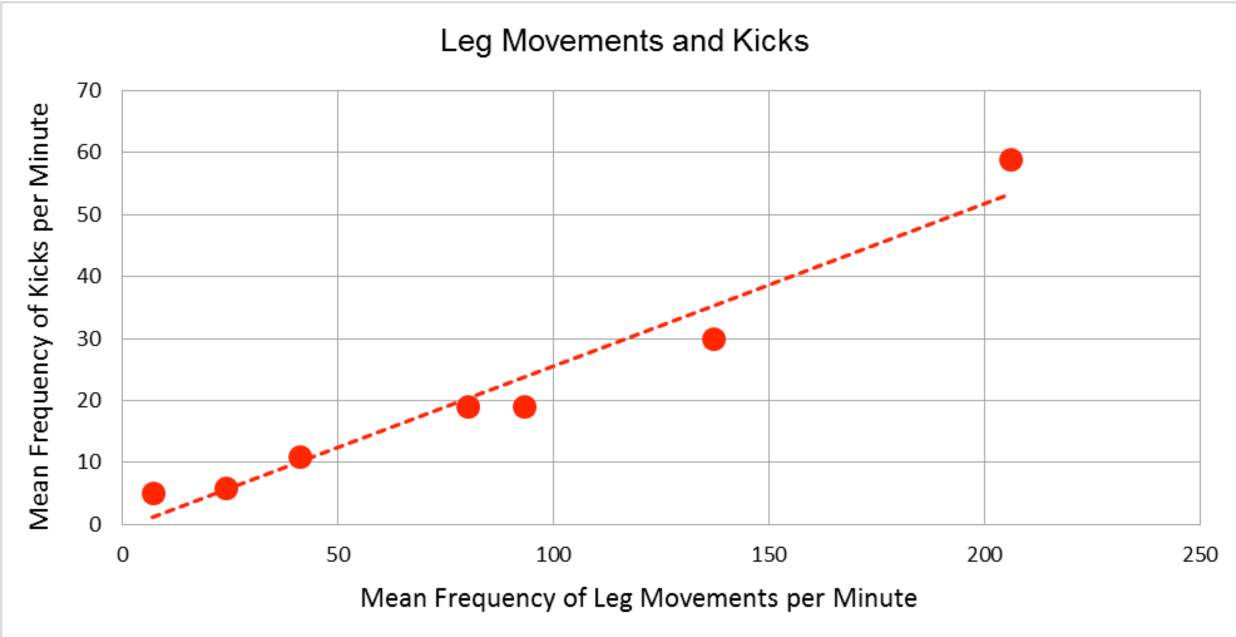


Figure 3. Relationship between the average number of leg movements and kicks generated by each infant across conditions

Figure 4 illustrates the significant difference observed between the mean frequency of tethered leg movements compared to the infant's untethered leg during the acquisition condition which was confirmed with a paired t-test {t (6) = 2.683, p = 0.036}.

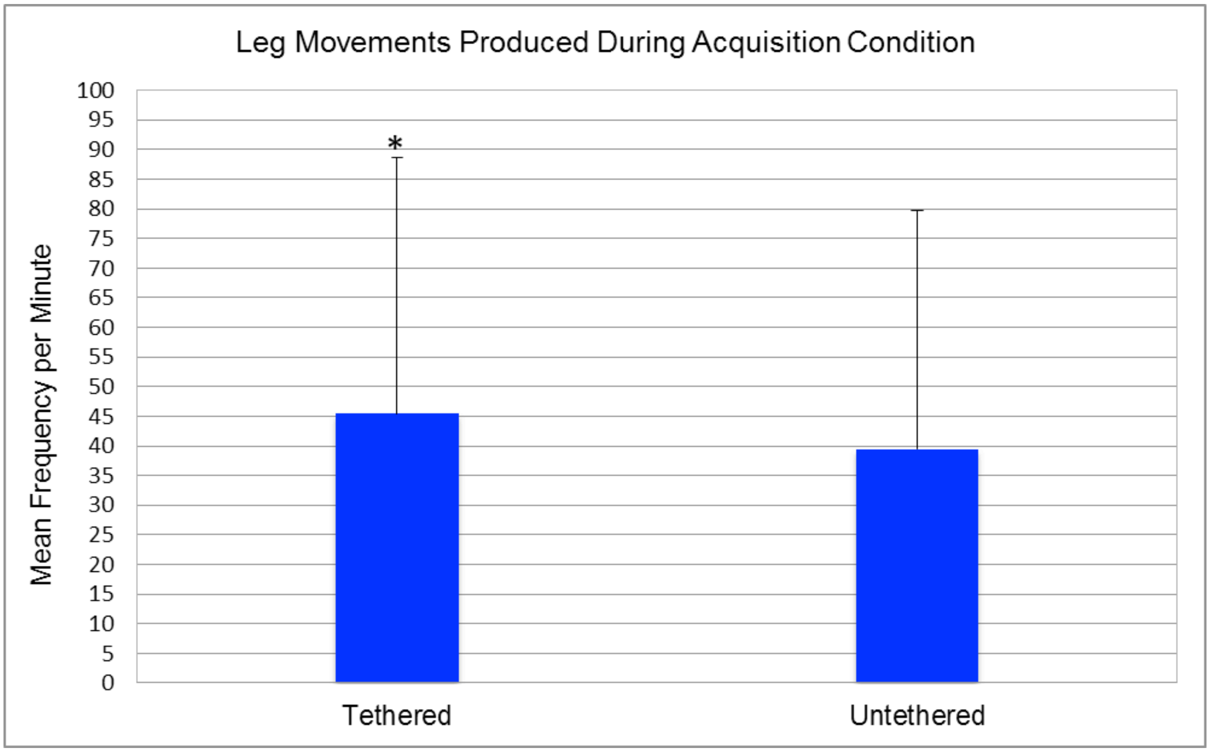


Figure 4. Tethered and untethered leg movements produced during the acquisition condition

Figure 5 displays the average number of spontaneous and goal-directed leg movements and kicks generated by this group of infants. A paired t-test was utilized to compare the frequency of spontaneous and goal-directed leg movements and kicks generated by this group of infants with SB. The highest frequency in the acquisition or extinction conditions was used for goal-directed leg movements and kicks based on previous studies that established this convention. In spite of the large standard deviations noted for each behavior, a significant difference was found between the number of spontaneous and goal-directed leg movements produced in either the acquisition or extinction conditions and baseline {p = 0.021}. The difference for kicks between the acquisition or extinction conditions and baseline only approached significance {p = 0.058}.

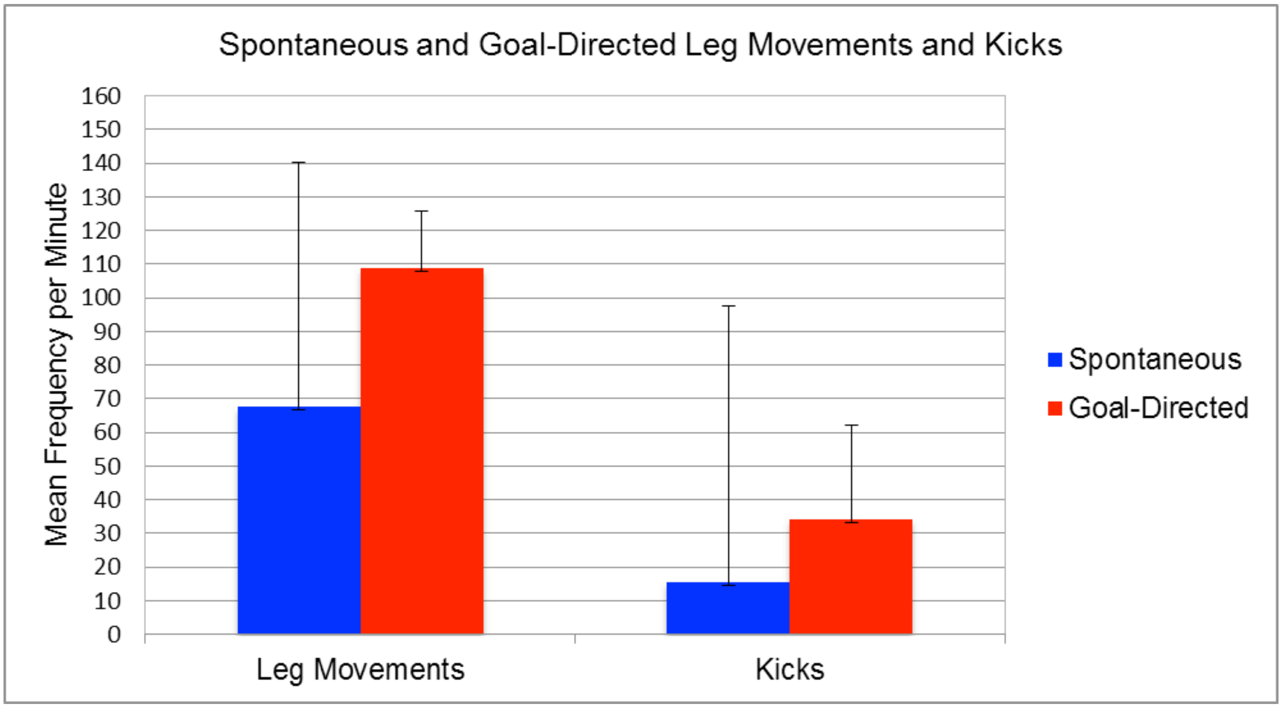


Figure 5. Frequency of spontaneous versus goal-directed leg movements and kicks

Figure 6 illustrates the frequency of leg movements for three exemplar babies in each condition who demonstrated low, moderate, and high activity levels, respectively. All three infants, regardless of their intrinsic activity levels generated more leg movements in the acquisition or extinction condition. The moderately active infant appears to have been most sensitive to the conjugate reinforcement paradigm compared to the low and high active babies given that he showed the greatest increase in leg movements as a percentage of his baseline leg movement frequency.

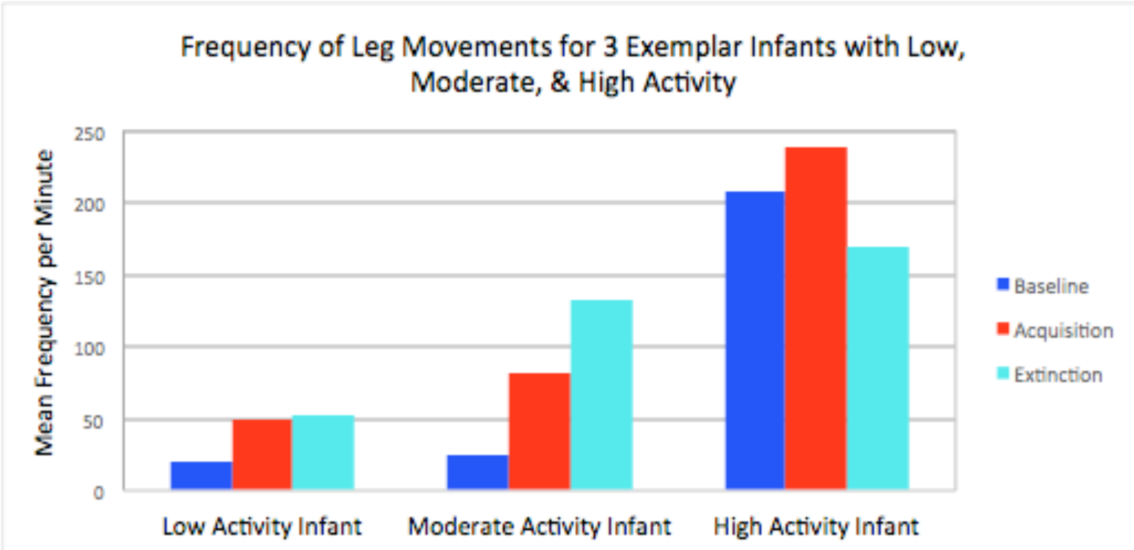


Figure 6. Frequency of leg movements for three exemplar infants with low, moderate, and high levels of activity

The frequency of kicks generated by the same three exemplar infants in each condition are presented in Figure 7. Similar to the data presented for leg movements these three infants also generated more kicks in the acquisition or extinction conditions compared to their baseline frequency of kicks. As with leg movements, the moderately active infant showed the greatest increase in the number of kicks generated per minute in the two experimental conditions compared to how often he kicked in the baseline condition.

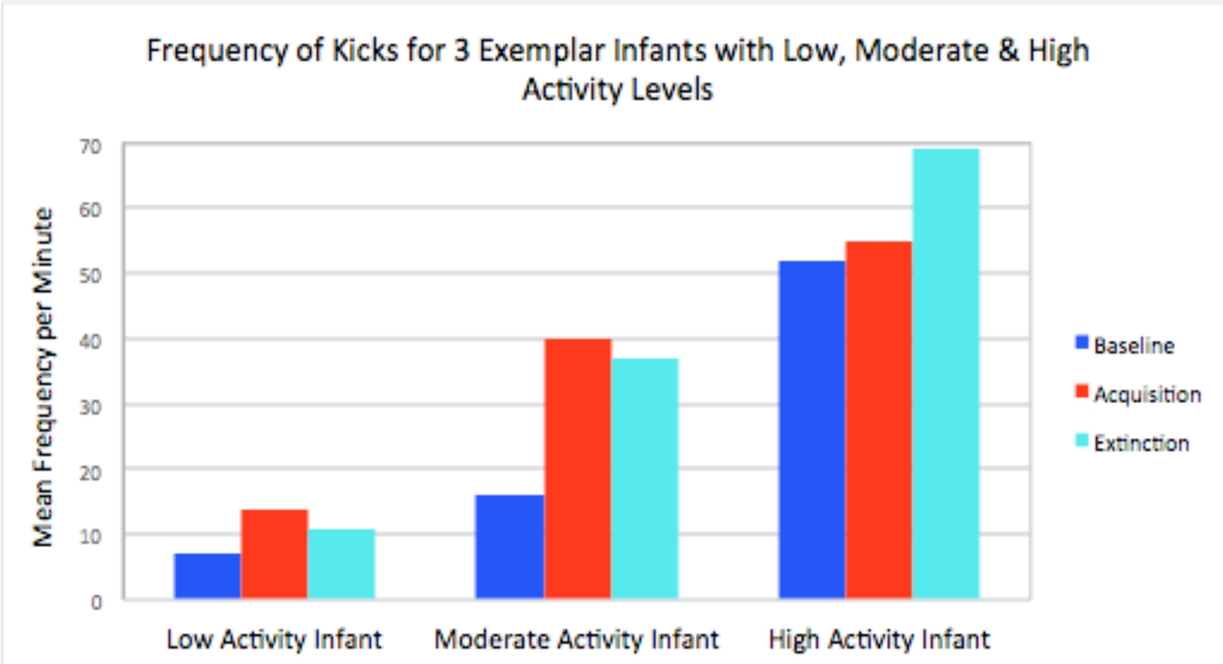


Figure 7. Frequency of leg movements for three exemplar infants with low, moderate, and high levels of activity

Three categories of kicks were behavior coded in order to analyze how often this group of infants with SB generated single, parallel, and alternating kicks. Figure 8 describes the mean frequency of each type of kick spontaneously produced in the BL condition and produced in a goal-directed condition of during the acquisition or extinction conditions. A one-way MANOVA was not significant, {$F(3,10) = 1.746$, $p = .221$} but this group of infants produced more spontaneous single kicks compared to spontaneous parallel or alternating kicks in the baseline condition. In general, the infants produced more of each kick type in the goal-directed paradigm compared to how often they kicked spontaneously in the baseline condition. Consistent with previous literature, the infants produced minimal numbers of alternating kicks in all three conditions.[30]

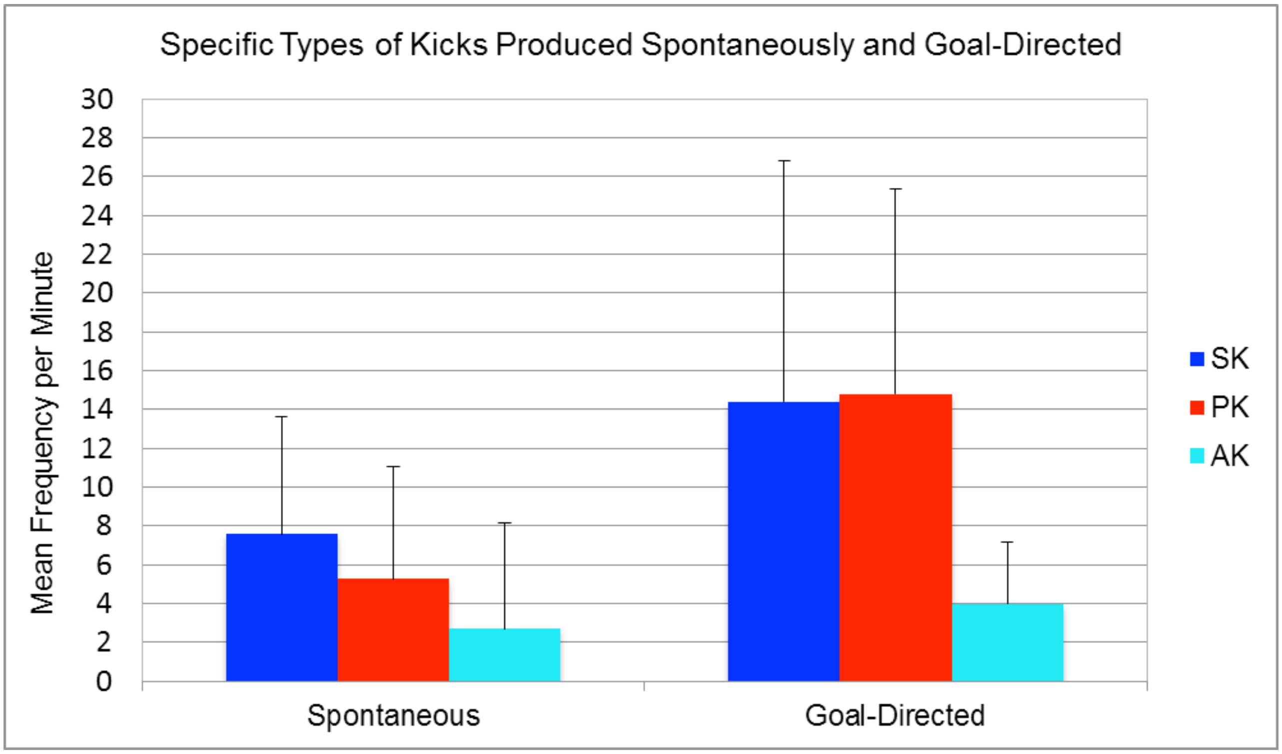


Figure 8. Average number of specific types of kicks produced spontaneously and in the goal-directed paradigm

Discussion

The purpose of this study was to examine the impact conjugate reinforcement has on the frequency of leg movements and kicks generated by infants with SB. We implemented a mobile paradigm and relied on the concepts of neuroplasticity and self-organization to create environment that helped infants with SB move their legs and kick more often that they spontaneously did in the BL condition. In light of the existing literature, our desire was to continue to discover techniques that may help infants with SB develop their motor skills.

As anticipated, we observed a significant, positive correlation between the number of leg movements and kicks generated by this group of infants with SB. The current results are consistent with a pilot study completed by Chapman et al that involved 3 babies.[30] We believe that this correlation will be useful to parents and other caregivers as well as clinicians. Parents and other caregivers can be encouraged to count their baby's leg movements rather than kicks which is an easier method for monitoring the activity level of their baby's legs. In clinic settings, therapists may choose to write treatment goals focused on facilitating leg movements rather than leg movements and kicks or just kicks knowing that there is a significant relationship between these two behaviors. In addition, an infant's lower extremity activity levels can be monitoredat home or in the clinic over time by counting leg movements rather than leg movements and kicks. The promotion of increased leg movements and kicks early in life is important for infants with SB as these early movement experiences may greatly impact when they achieve certain motor milestones, e.g. learning to walk.[31]

Although parents, caregivers and nurses often report that an infant with SB moves one leg more than the other we did not ask the parents or caregivers for this information prior to the study. The leg that was tethered during the acquisition condition was randomly assigned to avoid any activity level bias. The results showed that the tethered leg moved significantly more often than the untethered leg. This observation is consistent with the work of Rovee & Rovee and Collier who studied TD infants and infants with Down syndrome and Chapman's small pilot study (n=3) that was completed with infants with SB.[28,29,30] The observation that the tethered leg moved more often than the untethered leg suggests that if an infant demonstrates a difference in how often one leg moves compared to the other caregivers, parents and/or physical therapists may tether the less active leg in order to increase how often that baby moves her less active leg.[28,29,30]

Consistent with previous researchers who studied infants less than 12 months old with and with Down syndrome, a significant difference between the frequency of spontaneous and goal-directed leg movements using the conjugate reinforcement paradigm was observed.[28,29] This study's infants with SB were able to use visual and auditory stimuli in the goal-directed conditions to increase how often they moved their legs and kicked.

These results are important for several reasons. They extend the body of knowledge regarding how infants with SB move their legs and kick during the first year of life. The current data also provide additional support for using the concepts of neuroplasticity and self-organization to guide studies that examine the ability of infants with SB to move their legs and kick. The results also suggest that the conjugate reinforcement paradigm may be used to help infants with SB move their legs more often than they typically do. The mobile paradigm appears to create an environment in which an infant with SB may be able to move her legs more often which will strengthen the neural pathways that support leg movements and kicks in real time as she responds to the auditory and visual feedback provided by the mobile. This suggests that the conjugate reinforcement paradigm could be used to help infants with SB strengthen the leg muscles they use to move their legs and kick as well as increase the likelihood that they will move their legs and kick in the future.[21, 22]

This study was completed in light of the current literature, which confirms that infants with SB demonstrate fewer, less complex spontaneous leg movements and kicks when compared to TD infants. These behaviors are important to study in this population because infants who move their legs and kick more often walk earlier in life compared to their less active cohorts.[31] The delay in ambulation typically demonstrated by infants with SB limits their ability to explore their environment, which may impact their cognitive and social development as well as their ability to develop more complex movement skills like running and galloping.

We found that single knee and leg kicks were the most common type of spontaneous kick produced by this group of babies. However, in the goal directed conditions, this small group of babies generated slightly more parallel knee and leg kicks than single knee and leg kicks. And, like other groups of infants with SB, our group of babies generated very few alternating knee and leg kicks.[10, 13] We believe that infants with SB spontaneously generate fewer parallel and alternating kicks compared to how often they produce single kicks because these kicks are more complex. A single kick involves one limb and therefore at most two joints (hip and knee joints). In comparison, parallel and alternating kicks involve flexion and extension of both hips and knees, i.e. more degrees of freedom as well as the coordination needed to move both legs together or in alternation. As such, an infant must develop and implement a motor plan that involves an increased number of variables if they are going to generate parallel and alternating kicks.

In light of these observations, we believe that the conjugate reinforcement paradigm facilitates the ability of infants with SB to increase how often they generate complex kicks that require them to coordinate multiple joints and/or both legs. Based on the concept of neuroplasticity and the principle of self-organization, the conjugate reinforcement paradigm may be used to help infants with SB generate more complex kicks, i.e. parallel and alternating kicks, in real time as well as in the future. Thus, repeated exposure to the conjugate reinforcement/mobile paradigm may influence when they begin to walk.

Considerable variability for the frequency of spontaneous leg movements and kicks generated by this group of infants was observed in the BL condition. Despite these initial differences all of the babies increased how often they moved their legs and kicked in the goal-directed conditions. This observation suggests that it is important for clinicians, caregivers, and parents to assess the overall increase in leg movements from spontaneous to goal-directed rather than work to achieve a specific number in any given condition. Simply increasing how often an infant moves her legs provides her with greater opportunities to strengthen her leg muscles as well as the neural connections that support those behaviors. As a result, she will be more likely to move her legs in the future which may help her walk earlier in life than if she were not provided with additional opportunities to move her legs.[21, 31]

This study possesses several limitations. It relied on a relatively small sample (n=7). It is possible that a larger sample may yield different results. The infants who completed this study ranged in age from 16 - 42 weeks. A group of infants who were closer in chronological age may respond differently to the conjugate reinforcement paradigm than did our sample. A control group was not used. Thus, it was not possible to compare the response of TD infants to the conjugate reinforcement paradigm to how this small group of babies with SB reacted. The duration of trials were 2 minutes in length, which provides a small snapshot of how often the infants generate leg movements and kicks throughout the day. Shorter or longer trials may reveal a different response to the conjugate reinforcement paradigm. Only one of the infant's legs was tethered. The opposite leg was not tethered nor were both legs at once to see if that might impact how often the babies moved their legs. Finally, this study was a cross-sectional study rather than longitudinal. Therefore it is unknown how infants with SB will respond to this paradigm as they get older.

It would be beneficial to for future studies to use a larger sample size as well as involve younger babies. Younger participants, i.e. less than 16 weeks of age would allow researchers to see if this paradigm could be effectively introduced earlier in life. Future studies could also examine the impact that wireless motion sensors have on infant leg movements and if they are reliable in detecting leg movements. If the sensors are reliable, a parent or caregiver could be instructed to download their baby's leg movement data and a therapist could then analyze those results via ‘telemedicine’. It would also be beneficial to compare how TD infants, infants with SB who had their lesions repaired in-utero, and those whom had their lesions repaired postnatally respond to the conjugate reinforcement paradigm.

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