4-2015

The Effect of Conjugate Reinforcement on the Leg Movements of Infants with Spina Bifida

Sarah DeRosier
St. Catherine University

Jeremy Martin
St. Catherine University

Anna Payne
St. Catherine University

Kelly Swenson
St. Catherine University

Elisabeth Wech
St. Catherine University

THE EFFECT OF CONJUGATE REINFORCEMENT ON THE LEG MOVEMENTS OF INFANTS WITH SPINA BIFIDA

by
Sarah DeRosier
Jeremy Martin
Anna Payne
Kelly Swenson
Elisabeth Wech

Doctor of Physical Therapy Program
St. Catherine University

April 30, 2015

Research Advisor: Associate Professor David D. Chapman, PT, Ph.D

ABSTRACT

The Effect of Conjugate Reinforcement on the Leg Movements of Infants with Spina Bifida

Sarah DeRosier, Jeremy Martin, Anna Payne, Kelly Swenson, Elisabeth Wech

Advisor: David Chapman, PT, PhD

BACKGROUND AND PURPOSE: Current literature regarding the leg movements and kicks of infants with Spina Bifida (SB) is limited. Kicking reflects the motor development of the lower extremities, which influences the emergence of walking. The purpose of this study was to investigate the type and frequency of spontaneous and goal directed leg movements and kicks through the use of conjugate reinforcement in infants with SB.

METHODS: The spontaneous leg movements of five infants with lumbar or sacral SB were videotaped while supine: untethered (baseline); tethered to an overhead mobile (acquisition); and untethered (extinction). Data collection took place in each infant's home when the parents reported their baby was generally alert and active. Anthropometric measures were collected to determine if selected traits (eg thigh length) were related to the frequency of leg movements and kicks generated by the babies. Frame by frame behavior coding was used to identify leg movements and kicks.

RESULTS: All five infants moved their tethered leg more than their untethered leg in the acquisition condition. Participants generated significantly more leg movements in the acquisition or extinction condition compared to baseline ($p = 0.014$). The babies also generated more total kicks in the acquisition or extinction condition compared to baseline, but this was not significant ($p = 0.124$). Each baby responded uniquely to the conjugate reinforcement paradigm with respect to the frequency and types of kicks she generated (ie alternating leg kicks). There were non-significant, but moderate - high correlations between selected anthropometric measures (eg calf and thigh length, lower extremity range of motion) and leg movements and kicks.

CONCLUSION: These babies increased how often they moved their legs and kicked as a result of experiencing the mobile paradigm. Beyond frequency of leg movements and kicks, each baby demonstrated an individual response to the conjugate reinforcement paradigm by increasing the number of complex kicks she generated or demonstrating new types of kicks in the acquisition or extinction conditions compared to baseline. Clinically, the conjugate reinforcement paradigm provides babies with SB increased opportunities to strengthen their leg muscles and the neuromuscular connections that support kicks.

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

THE EFFECT OF CONJUGATE REINFORCEMENT ON THE LEG MOVEMENTS OF INFANTS WITH SPINA BIFIDA

submitted by
Sarah DeRosier
Jeremy Martin
Anna Payne
Kelly Swenson
Elisabeth Wech

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

Primary Advisor David Chen PT, PhD Date 4/30/15

Acknowledgements

We would like to thank the following for making this research project a successful and rewarding experience. First, we would like to thank the families of participants for welcoming us into their homes for our research. Secondly, we would like to thank Rachel Katoch, the research coordinator at Gillette Children's Hospital for assisting in locating the babies in our study. Third, we would like to thank David D. Chapman, PT PhD, our research advisor, for his guidance, leadership, expertise, and encouragement throughout the course of this research project. Lastly, we would like to thank our families and friends for their support during this project and our entire DPT graduate program.

Table of Contents

Introduction

In the United States about 1500 infants are born each year with Spina bifida (SB).[1] Despite being the most common neural tube defect in the United States, there is limited literature available regarding how infants with SB develop motor skills as well as the impact physical therapy may have on their motor development.

Spina bifida develops during the first month of gestation when the posterior aspects of the vertebral arches fail to properly fuse. This may allow the spinal cord and associated nerve roots to protrude dorsally exposing these neural tissues to potential injury.[2] Spina bifida lesions can develop at any level of the spine but most children have lesions in the lumbar or sacral regions.[3, 4]

There are multiple types of SB that present with varying degrees of severity. The mildest form of SB is SB occulta. It is characterized by a small under-developed gap in the vertebrae where the spinal cord is normal. It may be observable as a small hairy patch, dimple in the skin, or lipoma that lies over the bony malformation. Infants with this form of SB are typically asymptomatic.[1, 2] Spina bifida aperta accounts for 80% of SB cases and is further classified into meningocele or myelomeningocele.

Meningocele lesions occur when there is a large enough gap for meninges to protrude dorsally, but the spinal cord and nerves are unexposed and are intact. Individuals with this type of lesion may or may not have symptoms consistent with more severe SB cases. The most severe form is myelomeningocele where a sac containing the spinal cord and nerve tissue protrudes dorsally through the incomplete closure of the vertebral arches and is at risk to be injured.

Spina bifida can be diagnosed as early as 18 weeks of gestation through a maternal serum alpha-fetoprotein screen, ultrasonography, or amniotic fluid analysis. Risk factors that are linked to SB include genetic influences, exposure to teratogens, maternal use of anti-convulsant medication, and inadequate levels of folic acid intake.[1, 4]

Many infants with SB are delivered by cesarean section to decrease the amount of trauma associated with a vaginal birth and then have surgery to repair the lesion within 24-48 hours after birth.[5] In-utero surgery, which is a relatively new procedure, can be performed between 19 and 25 weeks of gestation. If the mother and fetus are recognized as candidates, general anesthesia is administered and an incision is made so that the fetus' back is exposed. The procedure then parallels the post-natal surgery where a pediatric neurosurgeon removes the sac and places a skin flap over the opening. This surgery aims to decrease the duration of exposure of neural tissue in the womb by removing the sac and placing a skin flap over the dorsal opening. Other proposed benefits of this surgery include a decreased need for a ventricular-peritoneal shunt, decreased severity of hindbrain herniation, and improved motor development.[5]

Infants with SB may experience a range of motor, sensory, or cognitive impairments, as highlighted in Table 1. Motor impairments such as scoliosis and clubfoot, motor paralysis, and spasticity are common. Visuoperceptual deficits, cranial nerve palsies, and neurogenic bowel and bladder incontinence are common sensory impairments. Babies with SB often have hydrocephalus and may also experience some difficulty with learning.[2, 6]

Table 1. Common Impairments Associated with Spina Bifida

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 incontinence	
Spasticity		

Infants with SB often experience delays in their motor development compared to infants who are typically developing. For example they learn to stand and walk much later in life, as highlighted in Table 2. Historically, the delays infants with SB experience in their motor development have been attributed to the level of their spinal lesion. That is, babies with higher spinal lesions generally demonstrate new motor skills later in life than babies with lower spinal lesions.[2, 6]

Table 2. Developmental Milestones

Developmental Milestone	**Typically Developing Infants**	**Infants with Spina Bifida**
Head Control	3 months	-----+
Rolling	4 months	-----+
Sitting	6-7 months	1-2 years
Crawling	7-11 months	1-2 years
Standing	9-13 months	3 years
Walking	12-14 months	3-7 years

-----+ Note: Ages for these milestones are not documented in the Literature.

More recently, researchers and therapists have begun to use the concept of neuroplasticity, which is the ability of neurons to change their function, chemical profile, or structure, as well as the principle of self-organization from dynamic systems theory to examine how and when infants with SB acquire new motor skills.[7] Self-organization means that babies learn how to coordinate their movements as a result of the interactions that occur between their sub-systems, e.g. muscles, ligaments, tendons, peripheral nerves and CNS with the environment(s) in which they are placed. This was first supported by Thelen's work where she held infants who no longer demonstrated an infant stepping response in chest deep water. She found that infants who were no longer able to generate an infant stepping response when held on a firm table top were able to generate steps when placed in a buoyant environment where they could overcome the effect of gravity.[8] This challenged the neuro-maturation approach, which suggests motor skills emerge according to a prescribed or hard wired neural template. If the neuro-maturation approach was completely true, the infants should not have been able to produce alternating steps in either of the environments.

Additional support for the principle of self-organization is provided by Chapman's work.[9] Guided by the principle of self-organization, he has shown that infants with lumbar or sacral SB are sensitive to their position in space and will alter how often they move their legs and kick between 4 and 13 months of age when they are placed in a specially designed infant seat compared to when they are supine or seated in a conventional infant seat.[9] Proponents of this approach suggest that over time and with repeated experiences, infants strengthen the muscles they use for particular movements (in this case, leg movements and kicks) as well as strengthen the neural connections that support their ability to move their legs and kick. These observations suggest that the environment can be used to facilitate or inhibit infant motor development.[9]

In this study, we focused on the frequency of leg movements and kicks because infants with SB usually have a decreased frequency of spontaneous leg movements and kicks when compared to their typically developing cohorts.[9, 10] In addition, they typically experience a delay in when they learn to walk compared to typically developing (TD) babies and usually demonstrate less frequent complex leg movements like leg and knee kicks.

As such, leg movements and kicks are important behaviors to study as they reflect the level of development and coordination of the lower extremities. Kicks are thought to be more complex than generalized leg movements. In particular, the type or types of kicks an infant demonstrates reflects the complexity of movement patterns they are able to coordinate. For instance, a kick involving only the knee joint is not as complex as one that involves the hip and knee joints. Parallel leg kicks, i.e. when the baby flexes and extends his or her legs at the hips and knees are perceived to more complex than a single knee kick, but less complex than alternating leg kicks in which the baby flexes and extends his or her legs at the hip and knee in alternation. Alternating kicks closely resemble the kinematic movement pattern used when we walk. Also, how often a baby moves his or her legs and kicks is related to when he or she will walk. Babies that move their legs and kick more often tend to walk earlier in life than infants who move their legs and kick less often during infancy.[11]

Previous research has focused on the ability of infants with lumbar and sacral SB to produce spontaneous leg movements and kicks. To date, no one has examined goal directed leg movements in infants with SB. Goal directed leg movements are considered to be leg movements that are intentional. As such, we chose to examine the impact of conjugate reinforcement on the leg movements and kicks in infants with SB. Conjugate reinforcement provides a reinforcing event which is a direct consequence of particular behaviors. So, a more frequent response produces a more intense reward value. For instance, as an infant moves his or her legs or kicks an overhead mobile will move and the infant may learn that his or her kicking is making the mobile move.

This paradigm was first implemented by Rovee and Rovee who tested 18 healthy infants ranging in age from 9-12 weeks in their home cribs.[12] They suspended an overhead mobile with hanging colored figures over the infant's head. A soft, silk ribbon was looped around the ankle of the infant and then tautly hooked to a suspended bar. Conjugate reinforcement was provided by means of the ribbon since foot and ankle movements directly initiated movement of the wooden figures. The experimental group had a baseline phase, an acquisition phase with conjugate reinforcement, and an extinction phase. The control group had the same three phases, but during the acquisition phase the experimenter constantly moved the mobile figures. They found that the operant response tripled within six minutes of the experimental group starting conjugate reinforcement while the control's response did not alter during the session. The infants in the experimental group learned the association between their movements and the movements of the mobile. This study demonstrated that intentional control of leg movements is already in place by three months of age in typically developing infants.[12]

The use of conjugate reinforcement to impact goal directed leg movements has also been examined by Collier who compared TD babies and babies with Down syndrome.[13] He used a mobile in a similar manner to Rovee and Rovee, which provided visual and auditory sensory stimulation in response to leg movements. He found that both groups of infants moved their legs more with reinforcement from the mobile and additionally moved their tethered leg more than their untethered leg.[13]

The mobile paradigm allows us to incorporate the concept of neuroplasticity and the principle of self-organization as we seek to discover new strategies that will enable babies with SB to move their legs and kick more often than they do spontaneously. We propose the mobile paradigm, which is a change in the infant's environment may provide them with opportunities to increase how often they move their legs and kick through goal directed activity. As a result, the purpose of this study was to investigate the frequency and type of spontaneous and goal directed leg movements and kicks through the use of conjugate reinforcement in infants with SB.

Methods

Participants

Prior to participant recruitment, Institutional Review Board approval was obtained. The participants were five full term infants, one boy and four girls, with lumbar or sacral SB. They ranged in age from 20 to 36 weeks at the time of data collection. Table 3 presents a summary of characteristics for each participant. All were recruited through the SB clinic at a large metropolitan hospital in the upper midwest. Participants received a $10.00 gift card as a participation incentive.

Table 3. Infant Characteristics

Characteristic	Baby 1	Baby 2	Baby 3	Baby 4	Baby 5
Age at Data Collection	5 months, 1 week	8 months	9 months, 1 day	9 months, 3 days	8 months
Full Term	Yes	Yes	Yes	No	Yes
Delivery Method	C-section	C-section, breech	C-section, breech	C-section	C-section
Lesion Level	L5-S1	L1	L5-S4	L2	L1
Lesion Repaired	Day 1	Day 1	In-utero at 25 weeks	Day 1	Day 1
Medications that would Affect Movement	No	No	No	No	No
Past Medical History	-No Shunt	-Missing L1-5, sacrum, coccyx -Bilateral club foot requiring bilateral tendon release, serial casts for 7 months, AFOs -No shunt	-No shunt -In NICU 3.5 weeks -Right club foot with AFO	-Bilateral club foot -Bilateral hip dysplasia, -Chiari Malformation	-V-P Shunt -Right hip dislocation -Left hip subluxation

Movement Data Collection

Data was collected in the baby's home at a time when the parent(s) reported that their baby was usually alert and active. Prior to video-taping each infant, a 0.5-meter long calibration rod was placed within the space the infant would occupy, parallel to the floor, and was video-taped for one minute. Then, the baby's clothes were removed down to his or her diaper to allow for maximum freedom of movement. Next, a small reflective hemispherically shaped marker (approximately 1.25 cm in diameter) was placed on the plantar aspect of the foot at the head of the first metatarsal in order to aid in later data reduction.

The baby was then placed supine on a towel on the carpeted floor of the parents' home. Next, a silk ribbon was tied around one of the infant's ankles. Randomization of right or left ankle was used to prevent any potential leg bias. Next, the mobile was positioned approximately 12-14 inches above the infant's head so that the mobile was out of the baby's reach. The mobile was a commercially produced infant mobile that is depicted in Figure 1. It is black and white, with four heart shaped stuffed cushions. Similar to previous research, four small jingle bells were also attached to the underside of the heart shaped cushions. As demonstrated in Rovee and Rovee when the child moved his or her legs or kicked while tethered to the mobile, the mobile would move, providing a visual and auditory stimulus to reinforce the baby for moving his or her legs and/or generating kicks.[12] During data collection, the parent(s), and/or student research assistants were seated near the infant but out of the infant's direct line of sight relative to the mobile.

Figure 1. A sample data collection set-up.

The leg movements of each participant were videotaped with a Sony Handycam that was positioned at a 23 degree angle relative to the floor and 1.5 meters, on average, away from the baby depending on the size of the living room. Camera placement was optimized for visualizing the subjects' lower extremities to aide data reduction. Two-dimensional data was collected rather than three-dimensional data as we were interested in capturing how often the babies moved their legs and kicked rather than the torques of their leg movements and kicks.

Based on the work of Rovee and Rovee[12] and Collier[13] the leg movements of each participant were videotaped at a rate of 30 frames per second in each of three conditions: baseline (BL), acquisition (ACQ), and extinction (EXT). The BL condition was two minutes long during which one of the baby's legs was tethered to the mobile stand so that the mobile did not move. Therefore, the leg movements generated in this condition were spontaneous leg movements. The ACQ condition was three minutes during which the same leg was tethered to the mobile to allow for leg movements and kicks that resulted in movement of the mobile. Therefore, the leg movements generated in this condition were goal directed movements. The EXT condition was two minutes long during which the same leg was again tethered to the mobile stand. Despite the leg being tethered to the mobile stand, the leg movements generated in this condition were again goal directed because the baby had experienced the ACQ condition and the opportunity to affect the mobile's movements. There was a short rest break between each condition. Trial lengths, i.e. two minute trials for the BL/EXT conditions and three minute trials for the ACQ condition, were based on earlier studies[12, 13] and ensured that a minimum number of leg movements were captured and to allow sufficient experimental impact for each participant.

Anthropometric Data Collection

After the movement test trials were completed, anthropometric measurements were taken by the faculty advisor for this study to examine the possibility that selected physical traits were related to the amount and type of leg movements observed. A description of each measurement taken can be seen in Table 4. The measurements included: total body length and weight; thigh and calf lengths, skinfolds, and circumferences; abduction of the right hip and right ankle dorsi-and plantar-flexion. Total body length and weight provided a measure of overall size. It was anticipated that heavier infants and those with larger skinfold measures and/or greater leg circumferences would move their legs and kick less often than leaner babies who had thinner legs. We also expected that infants with longer leg segments would move their legs and kick less often than infants with relatively shorter leg segments. The range of motion variables were measured to determine if infants with increased joint laxity moved their legs more or less often than babies with less mobile joints.

Table 4. Anthropometric Measures

Anthropometric Variable	**Measurement Technique**
Total Body Length	Baby is supine, looking up, body aligned with ankle dorsiflexed to 90 degrees. Measurement is taken from the top of the head to the bottom of the foot and recorded in inches to the nearest 1/16th of an inch
Thigh Length	The distance between the greater trochanter and the lateral condyle of the femur with the leg extended and the ankle dorsiflexed to 90 degrees
Calf Length	The distance between the lateral condyle of the femur and lateral malleolus with the leg extended and the ankle dorsiflexed to 90 degrees
	NOTE: Both leg length measures recorded in centimeters and measured to the nearest millimeter
Weight	Measured in pounds and ounces to the nearest ounce
Thigh Circumference	The circumference of the thigh measured at the midpoint of the segment length as measured above

Calf Circumference	The circumference of the calf measured at the midpoint of the segment length as measured above
	NOTE: The circumference measures recorded in centimeters and measured to the nearest millimeter and taken on the right leg
Medial Thigh Skinfold	The skinfold is vertical and taken at the same level as the thigh circumference on the medial surface with the thigh extended
Medial Calf Skinfold	The skinfold is vertical and taken at the same level as the calf circumference on the medial surface with the calf and knee extended
	NOTE: The skinfold measures recorded in millimeters and measured to the nearest .5mm and taken on the right leg
Hip Abduction	The infant was supine, with both legs extended at the hips and knees. The right hip was abducted
Ankle Plantar / Dorsiflexion	The infant is supine with the leg extended at the knee as the ankle is moved into plantarflexion and dorsiflexion
	NOTE: The three range of motion measures recorded to the nearest degree and taken on the right lower extremity

Data Reduction

The videotaped data was then behavior coded through a frame by frame analysis to identify the frequency of total leg movements, total number of kicks, and the frequency of nine types of kicks. The onset and end of leg movements were defined as occurring when movement of the marker attached to the bottom of the first metatarsal of the foot stopped, started or when a change in direction occurred.[3] For example, if a baby moved his or her leg medially then reversed direction and moved the leg laterally, the first movement ended and a second began at the point of change in direction.[9] Student researchers reached a percent agreement with an expert rater of $\geq$ 80% prior to reducing any data. After each trial was coded, the number of leg movements per minute and type of kick per minute was calculated.

Based on previous research, six types of kicks were identified through behavior coding of the videotaped data for each baby. These included single, parallel, and alternating leg kicks, which involved hip and knee flexion and extension. Single, parallel, and alternating knee kicks, which involved flexion and extension of the knee were also identified. However, because of the way the babies responded to the mobile paradigm, we are introducing the idea of a new category of kick: the hip kick, which involved flexion and extension of the leg at the hip joint. Hip kicks, like knee and leg kicks, were coded to reflect single, parallel, and alternating flexion and extension of the hip(s).

Data Analysis

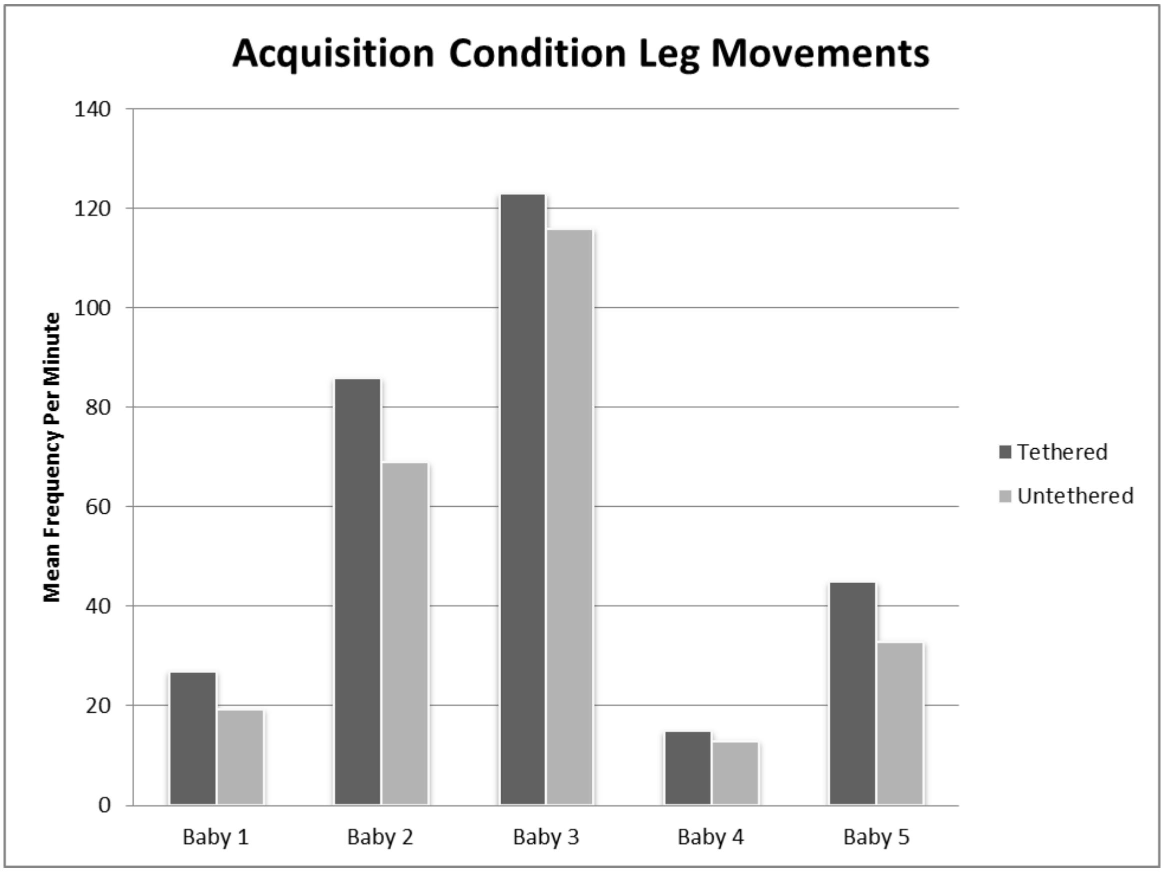
Subsequently, a paired t-test was utilized to determine presence of a significant difference in the frequency of the tethered and untethered leg movements in the ACQ condition. T-tests were also utilized to determine presence of significant difference in leg movements between BL and the highest frequency condition (ACQ or EXT). In addition, t-tests were used to confirm or deny the presence of significant difference in total kicks between BL and the highest frequency condition (ACQ or EXT). Lastly, the anthropometric measures taken were correlated with the frequency of total leg movements and kicks to determine the relative strength between the anthropometric measures and the infants' frequency of leg movements and kicks. All significance levels were established *a priori* at $p \leq .05$.

Results

Leg Movement and Kick Data

Figure 2 illustrates that each baby moved his or her tethered leg more than his or her untethered leg during the acquisition condition. As a group, this small set of infants with SB generated significantly more leg movements with the tethered leg compared to the untethered leg during the ACQ condition {t (4) = -4.013, df =4, p = .016}. These are results are consistent with previous research and highlight the impact that tethering one leg to an overhead mobile has on the frequency of leg movements.[13]

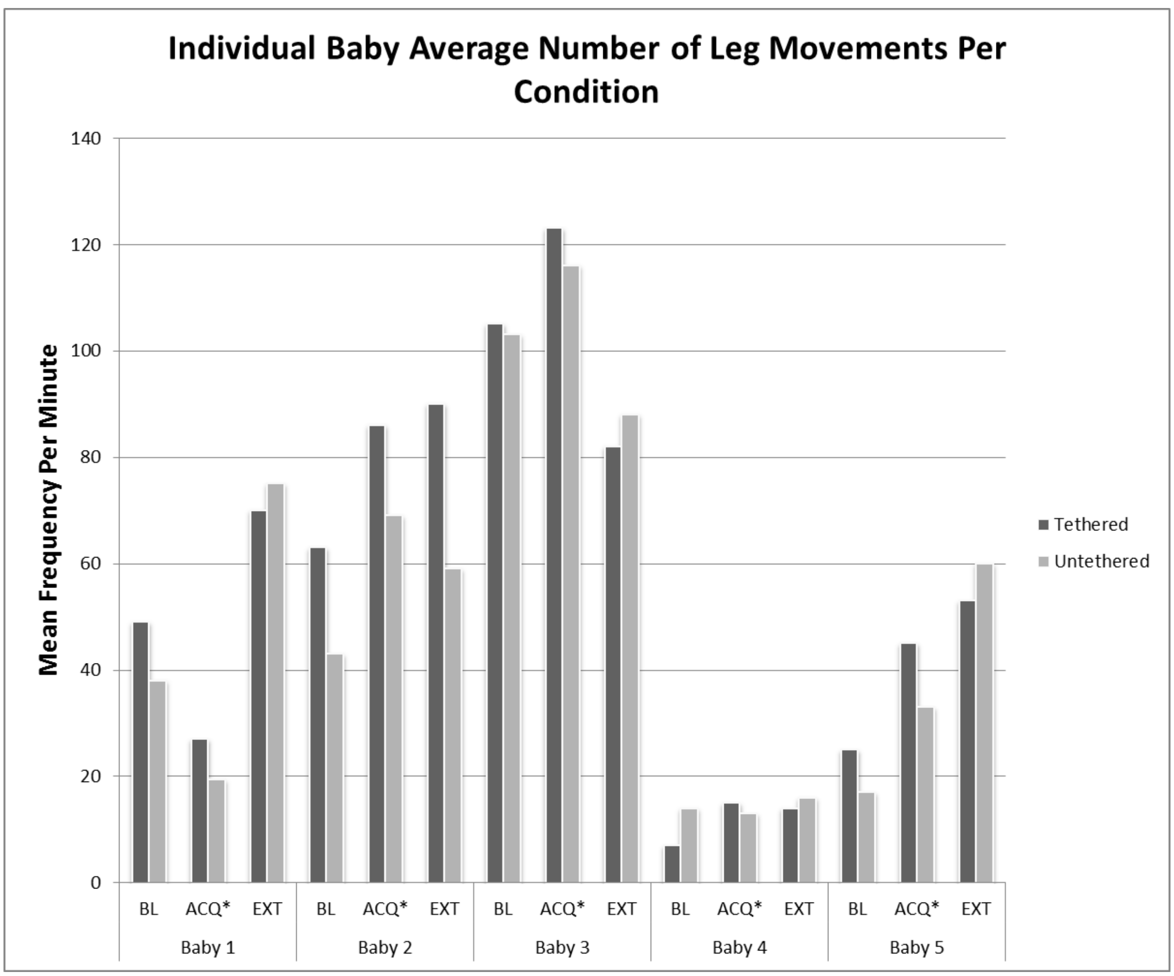
Figure 2. The average frequency of tethered and untethered leg movements generated during the acquisition condition.



The average number of leg movements produced by each baby in each condition are shown in Figure 3. Each baby responded individually to the conjugate reinforcement paradigm. Babies 1 ,4, and 5 moved their legs more in the EXT condition while babies 2 and 3 produced more leg movements during the acquistion condition compared to the EXT condition. We were most interested in deteremining if conjugate reinforcement assists babies in moving their legs more at any time after being tethered to the mobile. As a result, we perceived any increase in leg movements generated during the acqusition or

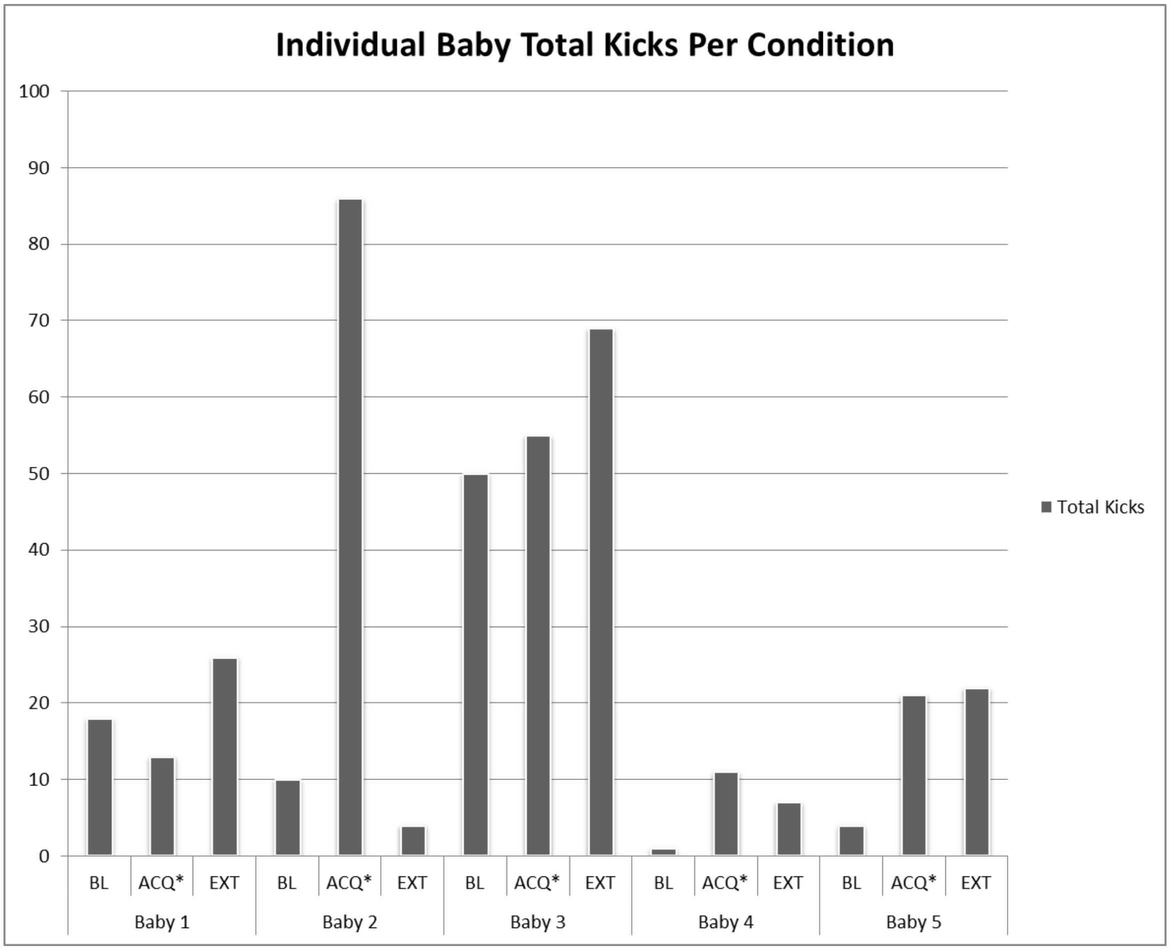
EXT condition to be beneficial. Thus, we used paired t-tests, conducted with the mean frequency of leg movements generated in BL and in the highest experiemental condition, i.e. ACQ or EXT for each infant, to confirm or deny a signficant condition effect on the average frequency of leg movements generated in each condition. This analysis revealed that the babies generated an average of 41 more leg movements per minute in the experimental conditions compared to BL, which was significant {t (4) = -4.131, p = .014}.

Figure 3. The average number of leg movements generated per minute by each baby in each condition.



The average number of kicks produced by each infant in each condition is presented in Figure 4. Because kicks are a subset of each infant's total leg movements we were not surprised to see that the babies generated fewer kicks than leg movements in each condition. Similar to what we observed for leg movements, each infant responded individually to the conjugate reinforcement paradigm. As Figure 4 depicts, babies 2, 3, and 4 generated the most kicks per minute in the ACQ condition while babies 1 and 5 did so in the EXT condition. Paired t-tests for kicks did not showed that these differences were not significant {$t(4) = -1.94$, $p = .124$}.

Figure 4. Average number of kicks generated per minute in each condition by each baby.



We also wanted to examine the impact conjugate reinforcement may have on the kick repertoire of these infants. For example, did the overhead mobile enable the babies to generate more parallel or alternating knee or leg kicks compared to when they were in BL? Exemplars of two infants' kick repertories are presented in Figure 5.

Figure 5. The kick repertoire of two exemplar babies.

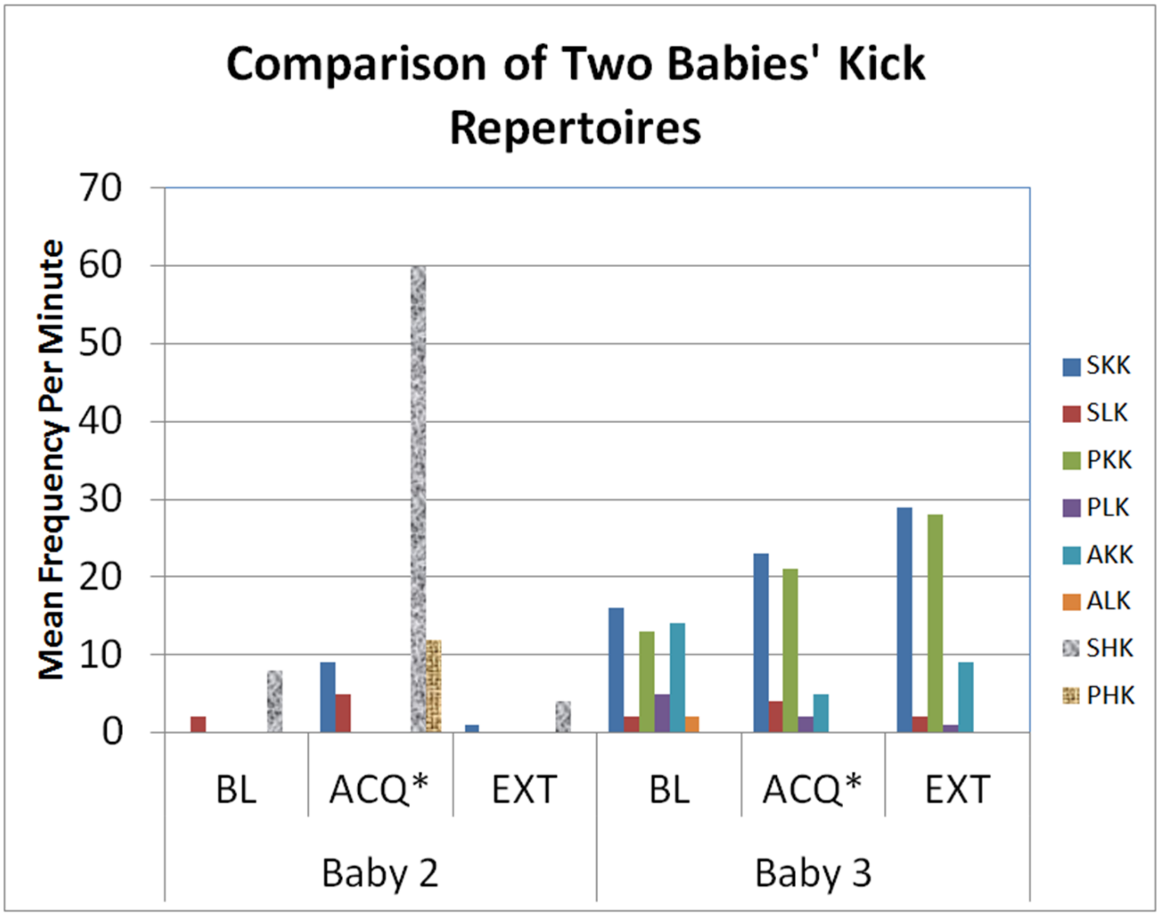


Figure 5 highlights the relative impact conjugate reinforement has on two of the babies in our study. Baby 3 had her spinal lesion repaired in-utero and was able to demonstrate a kick repertoire that is similar to what would be observed for a TD infant. She clearly demonstrated more single, parallel and alternating knee kicks compared to her peers and did not generate any hip kicks. Alternatively, Baby 2 displayed a relatively impoverished kick repertoire that was dominated by single and parallel hip kicks. Hip kicks have not been previously identified in the literature. Thus, we operationally defined a hip kick as occurring when the infant demonstrated flexion and extension at only the hip joint with no flexion nor extension being observed at the knee joint. Hip kicks seem to be relatively uncommon, but it appears to be related to the fact that this infant spent several months in serial casts to treat her bilateral club feet. As a result, she had not yet discovered her knee joint motions at the time of data collection.

Anthropometric Data

The relationship between the anthropometric data collected and total leg movements and kicks generated by this group of infants is presented in Tables 5-7.

Table 5. Correlations between leg movements, kicks and length variables

	Height	Thigh Length	Calf Length
Leg Movements	R = -.22, p = .722	R = -.562, p = .324	R = -.740, r = .153
Kicks	R = -.214, p = .73	R = -.387, p = .52	R = -.667, p = .218

Table 6. Correlations between leg movements, kicks and mass variables

	Weight	Thigh Circumference	Thigh Skinfold	Calf Circumference	Calf Skinfold
Leg Movements	R = -.338, p = .569	R = -.446, p = .452	R =-.645, p = .240	R = -.367, p = .544	R = -.106, p = .866
Kicks	R = -.172, p = .782	R = -.279, p = .649	R = -.491, p = .401	R = -.309, p = .612	R = .179, p = .774

Table 7. Correlations between leg movements, kicks and range of motion variables

	Plantarflexion	Dorsiflexion	Hip Abduction
Leg Movements	R = -.302, p = .621	R = .193, p = .756	R = -.373, p = .537
Kicks	R = -.445, p = .453	R = .245, p = .691	R = -.328, p = .59

These tables show that the relationship between the anthropometric data we collected with leg movements and kicks varied from small and positive, e.g. dorsiflexion with leg movements to strong and negative, e.g. calf length with leg movements. But, none of them reached or even approached significance.

Discussion

Guided by the concept of neuroplasticity and the principle of self-organization, we designed this study to investigate the frequency and type of spontaneous and goal directed leg movements and kicks through the use of conjugate reinforcement in infants with SB. We found that babies with SB, consistent with the literature for TD infants and infants with Down syndrome, moved their legs and kicked more often in the ACQ and EXT conditions compared to the BL condition when they were not tethered to the overhead mobile.[12, 13]

In our first hypothesis, we predicted that the babies would move their legs and kick more often in the ACQ or EXT conditions compared to BL. All of the infants demonstrated more leg movements and kicks in either the ACQ condition (when they were tethered to the mobile) or in the EXT condition (untethered), which took place immediately following the ACQ condition. This implies that infants with lumbar or sacral SB are sensitive to their movement context and are capable of generating goal-directed leg movements and kicks. Thus, the overhead mobile can be used to increase the frequency of their leg movements and kicks.

Several of our babies also demonstrated a greater array of kick types during the ACQ or EXT conditions compared to before they experienced being tethered to the overhead mobile. For example, Baby 2 demonstrated four kick types in the ACQ condition (single knee, single leg, single hip, and parallel kick) opposed to only two kick types in the BL condition (single leg and single hip). This suggests that the mobile paradigm may be employed to help infants with lumbar or sacral SB develop a richer leg movement and kick repertoire. This will enable them to strengthen their leg muscles and the neural connections that support leg movements and kicks.

In our second hypothesis, we predicted that the babies would move the tethered leg more often than the untethered leg. In our study, this hypothesis was confirmed, as all five babies moved his or her tethered leg more than his or her untethered leg during the ACQ condition. Our results are consistent with previous research that found that TD infants, as well as infants with Down syndrome, had a higher frequency of leg movements and kicks when one of their legs was tethered during the ACQ condition.[13] Similar to our findings, Rovee and Rovee reported that TD infants learned the association between their movements and the movements of the mobile.[12] Rovee and Rovee's study demonstrated that intentional control of leg movements is already in place by 3 months of age in TD infants and found that these babies moved their tethered leg more than their untethered leg. Collier's findings were also similar in that all of the infants in his study moved their legs more and, further, moved their tethered leg more than their untethered leg.[13] As anticipated, these babies did not move the leg that had been tethered more in the EXT condition when neither leg was tethered.

In our third hypothesis we anticipated that there would be an inverse relationship between mass and girth measurements with leg movements and kicks and that infants with longer leg segments would demonstrate fewer leg movements and kicks than infants with relatively shorter leg segments. None of the anthropometric variables (e.g. body weight, thigh, and calf length, skinfold, and circumferences etc.), were significantly related to how often the infants with SB moved their legs and kicked. Rather, there were varying relationships between the anthropometric data, leg movements and kicks. For example, dorsiflexion showed a low, positive correlation with leg movements while calf length demonstrated a high, but negative relationship with leg movements. These observations suggest that anthropometric measures do not seem to influence the spontaneous or goal directed leg movements and kicks of infants with lumbar or sacral SB. Our results are similar to previously completed research which found that there were not significant correlations between these types of anthropometric measures, leg movements, and kicks.[9] They are also consistent with previous reports that described varying levels of association between leg movements, kicks, and anthropometric data for infants with SB.[14,15] However, our study differs slightly from earlier work by Chapman who found a slight a trend for infants with heavier legs to move their legs and kick less than infants with smaller, lighter limbs.[9,14]

Our findings have several clinical implications. Caregivers should consider positioning infants with lumbar or sacral SB in ways that enable them to move their legs and kick as often as possible. Alternatively, parents and other caregivers should avoid contexts, like conventional infant seats, that inhibit the ability of infants with lumbar or sacral SB from moving their legs and kicking. The mobile paradigm which appears to facilitate the leg movements and kicks of infants with lumbar or sacral SB coupled with previous research strongly supports the idea that the frequency of leg movements and kicks influences how babies develop leg strength and neuromuscular control of their lower extremities.[9,10,11,12] In addition, infants with SB appear to be sensitive to visual and auditory feedback, both of which may be used to increase how often babies with lumbar or sacral SB move their legs and kick.

There are several limitations to our study. These include:

1. A small sample size (n=5)
2. The age range of the current participants (5-9.5 months)
3. Tethering only one leg
4. Length of the ACQ condition was limited to 3 minutes

5. Exclusion of TD infants
6. Cross-sectional design

Future research should strive to recruit additional participants and investigate tethering both of the infant's legs (one at a time in a randomized fashion). Future researchers may also investigate the frequency and repertoire richness of leg movements over time via a longitudinal design with the mobile paradigm. We suggest additional research to determine if increasing the frequency of early leg movements, via therapeutic intervention, will enable infants to increase the duration and frequency of spontaneous and, or goal directed leg movements and kicks. Coding for hip kicks should be continued in future studies. This was a novel kick identified for the first time in our study.

Conclusion

How often a baby moves his or her legs and kicks is related to when he or she will walk.[11] These infants with lumbar or sacral SB demonstrated more goal directed leg movements and kicks as a result of the mobile paradigm compared to their spontaneous leg movements and kicks. According to Rademacher, depressed activity levels may reflect inherent disruption to the neuromotor pathways which would reduce the infants' opportunities to learn to develop control of their lower extremity movements and strengthen the neuromuscular organization and cooperation needed to acquire functional motor skills, like pulling to a stand and walking.[10] Thus, if we can continue to discover and design these types of movement experiences we may be able to help infants with lumbar or sacral SB walk earlier in life than they currently do.

References

1. Centers for Disease Control. Spina Bifida. http://www.cdc.gov/ncbddd/spinabifida/data.html. Accessed Nov 2014.
2. Campbell SK, Palisano RJ, Orlin MN. Physical Therapy for Children. Saunders; 2012.
3. Chapman DD. Context Effects on the Intrinsic Dynamics of Infants with Spina Bifida (SB). Bloomington:Indiana University; 1997.
4. 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 Nov 2014.
5. Children's Hospital of Philadelphia. About Spina Bifida. http://www.chop.edu/conditions-diseases/spina-bifida/about#.VNLbdS4vvG4. Accessed Nov 2014.
6. Engstrom A, Lucken S, Sis K, Wehrheim S. The influence of position and age on leg movements and kicks in infants with spina bifida. St. Catherine University Research Day. Minneaplolis, MN. Dec 2013.
7. Lundy-Ekman L. Neuroscience, Fundamentals for Rehabilitation.Saunders; 2012.
8. Thelen E, Fisher D, Ridley-Johnson R. The relationship between physical growth and a newborn reflex. Infant behavior and development, 1984, 7(4):479-483.
9. Chapman D. Context effects on the spontaneous leg movements of infants with spina bifida. Ped Phys Ther. 2002;14(2):62-73.
10. Rademacher N. Early spontaneous leg movements in infants born with and without myelomeningocele. Ped Phys Ther. 2008;20(2):137-45.
11. Ulrich, BD & Ulrich, DA.Spontaneous leg movements of infants with Down syndrome and nondisabled infants. *Child Dev*. 1995; 66:1844-1855.
12. Rovee C, Rovee D. Conjugate reinforcement of infant exploratory behavior. Journal Of Experimental Child Psychology [serial online]. August 1969;8(1):33-39. Available from: MEDLINE, Ipswich, MA. Accessed February 6, 2015.
13. Collier D. Goal directed movement in infants with Down syndrome [dissertation]. Bloomington, IN; Indiana University, 1993.
14. Chapman D, Engstrom A, Lucken, S, Sis K, Wehrheim, K. *The Influence of Age, Position, and Timing of Spinal Lesion Repair on the Ability of Young Infants with Spina Bifida (SB) to Move and Kick Their Legs.* Paper presented at the Minnesota APTA Spring Conference; 2014. St Paul,MN.

15. Chapman, D. The Development of Leg Movements and Kicks in Older Infants with Spina Bifida. *Ped Phys Ther*. (In review)