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Behavior During Tethered Kicking in Infants with Periventricular Brain Injury

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Abstract

Purpose—Describe behavior of children with periventricular brain injury (PBI) in a tethered-kicking intervention.

Methods—Sixteen infants with PBI were randomly assigned to exercise or no-training in a longitudinal pilot study. Frequency of leg movements and inter-limb coordination were described from videos at 2 and 4 months corrected age (CA).

Results—Eight of 13 children (62%) with longitudinal data increased the frequency of leg movements while tethered to a mobile between 2 and 4 months CA. Movement frequency was correlated with scores on the Test of Infant Motor Performance, but there were no differences between experimental groups. Children with typical development at 12 months CA increased the proportion of leg movements that were synchronous between 2 and 4 months as did a child with cerebral palsy in the experimental group.

Conclusion—The tethered-kicking intervention facilitates movement in infants with PBI but effects on development remain to be demonstrated.

Children with periventricular brain injury (PBI) have high rates of poor developmental outcome ranging from 50–90 percent,[1,2] but little is known about differences between those who recover with typical development (TD) and those who go on to have atypical development such as cerebral palsy (CP) or delayed development (DD) of motor skills. Yang and colleagues documented the occurrence of many abnormal postures in children who developed CP, but only inability to maintain the head in midline alignment in supine was present in a majority (63%) of the children in their study.[3] Sixteen percent of subjects with CP showed a paucity of leg movements, while 18% had prolonged, monotonous kicking.[3]

Longitudinal observations of functional movements of children who recover from perinatal brain insults, i.e., do not develop CP or delay, are rare. We previously reported the effects of a 10-month-long task-oriented home-based kicking and stepping exercise intervention on overall gross motor development and walking at 12 months CA in a longitudinal pilot study with infants with PBI, half of whom were typically developing at 12 months CA.[4,5] No statistically significant effects on performance on the Alberta Infant Motor Scale were found, yet more children in the experimental group were walking alone or with one hand held at 12 months CA. Because the effects of the exercise intervention were unclear, we believed it was necessary to evaluate whether the exercise program we used was tolerated by the infants and feasible as a means of eliciting active leg movements as intended. Thus the present report is an analysis of results at the behavioral level from 2–4 months CA when experimental group children were randomly assigned either to exercise in a tethered-kicking program or no exercise. We aim to better understand the effects of age, developmental outcome, and the exercise intervention on leg movements, attention and head control.

Spontaneous kicking, i.e., hip flexion and extension movement, has been studied in a variety of infant populations. Infants with white matter damage of the brain have a higher spontaneous kick frequency than term or preterm infants without brain insults from 1–3 months CA, but no significant differences in the kinematics of kicking that are predictive.[6]

Infants with brain insults demonstrated more crying which was accompanied by higher kick rates. Jeng and colleagues reported that infants who later were found to have DD had more unilateral and fewer synchronous kicks at 2 months CA but the opposite at 4 months CA.[7] A higher kick frequency at 4 months CA predicted a later age of achievement of walking. Fetters and colleagues suggest that infants with brain insults move in a different, more extended, space with restricted range, less variability, and tighter coupling among leg joints at one month CA,[8] and at five months of age, they move into hip flexion from a less extended starting position at the hip and knee than infants born at term.[9] Decoupling of the movement of intra-limb joints is proposed to be a necessary developmental change in order for more complex movements to evolve.[10] This natural maturational process may be more difficult to achieve for infants with brain insults when functional movements, rather than spontaneous kicking, are attempted. To our knowledge there are no reports on the development of kicking as a functional activity, i.e., a purposeful, goal-directed activity, in infants with PBI.

The classical mobile paradigm involves measurement of purposeful leg movements with one leg tethered to a mobile such that kicking initiates movement of the mobile. In the typical study the focus of analysis is on the cognitive ability of the infant to recognize that the movements of the tethered leg activate the mobile. Heathcock and colleagues[11] described the physical, behavioral and perceptual systems required for performance in the classical mobile paradigm as: 1) arousal and self-regulation, 2) visual attention, and 3) ability to make spontaneous leg movements. Given the difficulty with maintenance of the midline head position identified in CP in the report of Yang and colleagues,[3]ability to control head position might be another prerequisite to successful performance in the mobile paradigm (if the head is not fixed in midline by the experimenter). Infants born at term can learn to activate the mobile in a single 10- to 15-minute session at 3–4 months of age. On the other hand, infants born preterm require significantly more time to learn that their kicking can effect movement of the mobile[11] and in general show less purposeful leg control.[12] Haley and colleagues[13] demonstrated that infants born preterm spend less time looking at the mobile, and have lower cortisol levels and more negative affect than infants born full term. To our knowledge, a mobile has not been used as a means to the end of facilitating leg movements in infants at risk for poor developmental outcomes as a result of PBI, but using a mobile to create an interesting visual and auditory response contingent on the infant's leg movements might fulfill the need for creating a task-oriented functional activity for the very young infant.

Despite the difficulty shown by infants born preterm in learning to control mobile movements, we believed that a tethered-kicking program might be a useful way to promote muscle activity and functional leg movements in children with PBI who have a high risk for developing DD or CP. Many of these children develop spastic diplegia with weakness in the leg muscles and postural abnormalities such as excessive hip adduction and internal rotation with plantar flexion of the ankle.[14] We supported each leg with an independent ankle sling tethered to the mobile, which was expected to facilitate freedom of movement for these children because the slings supported part of the weight of the legs. We expected that regular practice would promote midline head control, visual attention, and active use of the legs to make the mobile move. In this paper, we describe behavioral state, head control, kicking frequency and kicking patterns in a tethered-kicking program designed to promote motor development in infants with PBI between 2 and 4 months CA. Our goal was threefold: 1) to examine the feasibility of using a tethered mobile in an exercise program from 2–4 months CA to facilitate leg movements in infants with PBI, 2) to follow up on the possible effects of the intervention on movement frequency and patterns of leg movements after 2 months of exercise, and 3) to examine differences in tethered-kicking behavior between infants who recovered from PBI with typical motor development and those who had atypical development, i.e., CP or DD, at 12 months CA.

Methods

Subject Recruitment

The Institutional Review Board at each participating institution approved the protocol for this study, and we obtained parental assent for enrollment of subjects in the study. Infants were recruited from three neonatal intensive care units (NICUs). Infants with PBI were included if they had Grade III or IV intraventricular hemorrhage (IVH) or periventricular leukomalacia (PVL) as visualized on ultrasound scan and reported in the medical record during the perinatal period. Physicians in each NICU verified that subjects were expected to be healthy enough at discharge in order to begin a home exercise program at 2 months CA. A random numbers table was used at each site to assign subjects to experimental groups (kicking exercise using the tethered mobile or control). Only the principal investigator and, for infants in the exercise group, the physical therapist who taught the exercise program to the parents, were aware of the group assignment.

Intervention

Children assigned to the exercise group received monthly visits from a physical therapist who provided them with a mobile with Velcro tethers to attach independently to each ankle such that kicking or other leg movements rewarded the infant with interesting visual and auditory feedback for exercise from 2–4 months CA (Figure 1). Children were positioned in an infant bath seat for the tethered-kicking training. Therapists showed parents how to set up and use the mobile, explained the theory behind the kicking exercise, and reviewed the expectations for performing the exercise. Although parents were shown how the toys worked to produce interesting feedback and occasionally moved the infants' legs to encourage them to kick, the infants were allowed to explore and play with the toys without additional handling. Parents were asked to perform the kicking exercises for 8 minutes per day/5 days per week. Further details of the intervention program, which continued to 12 months CA with additional activities, were previously reported.[4] Children in the control group were visited only for assessment at 2 and 4 months CA. Other types of early intervention were allowed for either group.

Developmental Assessment

One of three physical therapists masked to experimental group assignment visited infants in both groups at 2 and 4 months CA for testing of motor development and videotaping of the infants' behavior while tethered to the mobile. Parents were asked to avoid revealing their group assignment during these visits but 7 parents inadvertently did so. This was a longitudinal study with motor development tested at 2 (study entry) and 4 months CA with the Test of Infant Motor Performance (TIMP)[15] and at 12 months CA with the Alberta Infant Motor Scale (AIMS).[16] At 12 months CA, all children were assessed by a pediatric rehabilitation medicine physician unaware of group assignment or AIMS performance until after the examinations were completed. After administering a standard protocol to assess reflexes, postural tone and movement quality, the physician judged whether the child did or did not have CP, and was then given information on the child's AIMS performance. The physician assigned those with CP a functional level based on the Gross Motor Function Classification System (GMFCS) categories for children before the 2nd birthday [www.canchild.ca].[17]

Assessment of Behavior While Tethered to the Mobile

Infants in both groups were videotaped in their home at a time when parents expected the infant to be awake for approximately 4 minutes with a single camera at both 2 months CA (baseline, no previous tethered-kicking experience) and again at 4 months CA while seated in the infant bath seat with ankles tethered to the mobile. Behavioral state, and head and leg movements were coded using the Datavyu Version 1.04 coding program [www.datavyu.org]. Behavioral state was coded in a first pass through the video as 1) awake, 2) asleep, or 3) crying. In a second pass through the video, head movements were coded as 1) midline if the infant appeared to be looking at the mobile, 2) right turn if the head and eyes moved toward the right away from the mobile, and 3) left turn if the head and eyes moved to the left away from the mobile. Only periods in which the infant's behavior was coded as awake were included in the final analysis of head and leg movements. In addition, periods when the experimenter was interfering with movement by adjusting the slings or passively moving the legs to encourage kicking were coded as segments to be removed from the final data analysis. Movements at the hip (flexion/extension) in each leg were coded in 4 separate passes through each video. The criterion for recording an event as a leg movement was that the hip was observed to move more than about 15 degrees in either direction from the resting position in the slings of approximately 90 degrees of hip and knee flexion. Movements coded were 1) right hip flexion (movement into hip flexion from the resting position), 2) right hip extension (movement extending the hip from the resting position), 3) left hip flexion, and 4) left hip extension.

Two physical therapists coded the video data to assess inter-rater reliability. Although both coders were aware that all infants in the study had PBI, the video was viewed and coded without knowledge of the infant's outcome or experimental group assignment. After coding by the primary analyst, a quasi-random selection of one minute of activity was prepared for analysis by the second coder. For the first subject, the first minute in which the infant was awake with no interference by the experimenter was selected, for the second infant the second minute, etc. Leg movements were considered to be in agreement if both coders marked the same movement event as occurring within 1 sec of each other. All disagreements were reviewed by both analysts. Final agreement averaged 83% across leg movement types at 2 months CA and 89% at 4 months CA. Finally, inter-limb coordination was recorded by the primary coder based on each flexion movement of the right hip according to the terminology of Jeng and colleagues[7] as 1) ipsilateral, 2) alternate (defined in this study as left leg extending within 1 sec. of right hip flexion), or synchronous (defined as both hips flexing within 1 sec. of each other).

Data Analysis

The design of this study was a longitudinal pilot study of an exercise program to facilitate motor development of infants with PBI. The level of analysis in this paper is the behavior of the infants while the ankles were tethered to the mobile, including behavioral state, head control, and leg movements. Time in the awake, alert state was calculated as the proportion of time awake divided by the total of time awake plus time asleep plus time crying. Head position in the awake state was calculated as the proportion of time with the head in midline divided by the total of time midline plus time with the head turned to the right or the left. Leg movement frequencies (movements/minute in awake state with no crying) were calculated separately for all four leg movements at 2 and 4 months CA, and the frequency of the total of all four movements/minute was obtained for each child. T tests were used to compare average movement frequency across leg movements between 1) infants with TD at 12 months CA and 2) those with CP or DD, which allowed us to include all children with data at either time point. Based on Bonferroni correction, the level of probability set for accepting statistical significance for multiple t tests was set at $p = .01$.

To evaluate longitudinal change in performance across time and with intervention, repeated measures analysis of variance (ANOVA) was used, which included only children with data at both ages (N=13). The ANOVA compared average leg movement frequencies at 2 and 4 months CA among 1) children with TD at 12 months CA, 2) children with CP, and 3) children with DD but no CP and, second, between experimental (tethered-kicking practice) and control groups. Proportions of right hip flexion movements that were synchronous (versus ipsilateral or alternate) were also compared among the 3 developmental outcomes and between 2 months and 4 months CA using repeated measures ANOVA.

Results

Subjects

Eighteen subjects with PBI were initially recruited from the three NICUs. One subject was withdrawn after parental assent was obtained but prior to beginning of data collection and training because the family changed their mind about participation; a second subject was too ill to ever begin study participation and died during the course of the study. Thus, 16 subjects randomly assigned to one of two experimental groups (7 exercise, 9 control) completed the study. No attrition occurred after any child began the exercise or testing program, although one subject could not be scheduled for the final AIMS test until 16 months CA. One child was not assessed at 2 months CA because she was ill and two children were not videotaped in the tethered-kicking setup at 4 months CA.

The sample consisted of 8 males and 8 females. The range of gestational age at birth was from 23–32 weeks with a mean of 27 weeks (SD 2.9 weeks, Table 1). Nine subjects were African-American, 5 white (one Middle-Eastern), and 2 Hispanic. There were no statistically significant differences between groups in gestational age at birth, type of brain insult, chronic lung disease, frequency of other physical therapy, or motor development at study entry.[4] Because children with PBI in Illinois are automatically eligible to receive early intervention services, we expected that all of the subjects in this study would receive additional physical therapy. In actuality, however, only 4 children (1 experimental, 3 control) were receiving physical therapy in the two months during which tethered-kicking exercise was provided to the infants in the experimental group.

Developmental Outcome

The exercise group (N=7) had 3 children with TD at 12 months CA (AIMS scores were at or above the 50th percentile with no abnormal neurologic findings) and 4 with atypical development: 3 with CP and 1 with DD (AIMS score < 10th percentile but no CP).[4,5] The control group (N=9) had 5 children with TD, 3 with CP and 1 with DD. Thus, there were more children in the (larger) control group with normal developmental outcome, but CP and DD were represented equally in the two groups. All children with CP or DD at 12 months CA also had delayed development (scores < −0.5 SD) on the TIMP at 4 months CA (Table 1).

Arousal and Self-regulation

At both 2 and 4 months CA, infants showed the ability to maintain an awake, alert state while sitting in the bath seat with ankles tethered to the mobile. Only four infants cried during the videotaping session at 2 months CA, 3 children with TD outcome at 12 months and one diagnosed with CP at 12 months. The child with CP cried for a total of 2.3 minutes while crying in the children with TD typically averaged less than a minute. In addition, one child with CP fell asleep for almost a minute at the end of the session. At 4 months CA, all children remained awake and alert throughout the videotaping session. One child, however, was observed to have a pacifier in her mouth throughout the 4-month session despite no apparent signs of distress. Although video segments in which a pacifier was used in response to infant fussing were otherwise not included in the analysis of kicking frequencies, in this case pacifier use was accepted rather than deleting the child’s data completely from the analysis.

Visual Attention and Head Control

At both 2 and 4 months CA, infants appeared to be highly engaged in the task, looking at the toys on the mobile and switching their gaze from one hanging toy to another. They also took “breaks,” e.g., looking at people in the room, before returning their attention to the task. The average proportion of awake time with the head in the midline was high (82–94% of the time) for all three outcome groups at both ages (Figure 2) except at 2 months CA for children with DD who averaged 54% (SD 31%) of the time with the head in midline. All children with TD maintained a midline head position averaging 93% (SD 12%) of the time at 2 months CA and 85% (SD 15%) at 4 months CA. Two subjects with CP had more difficulty maintaining the head in midline: Subjects E6 (Table 1) at 4 months CA (52%) and C7 at 2 months CA (65%). One subject with CP (C9) was later found to have visual impairment requiring glasses but averaged attention to the task with the head in midline of 81% at 2 months and 80% at 4 months despite frequent head turns to either side. Although children with DD or CP were able to engage in the task, they did have more difficulty maintaining the head in midline than children with TD.

Leg Movements While Tethered to the Mobile at Two Months CA

Mean frequencies of the four leg movements (right hip flexion, right hip extension, left hip flexion, and left hip extension/minute) and the means of total movement frequency are presented in Table 2, and data for each individual subject showing overall frequency of leg movements (sum of all 4 movements on both legs/minute) can be found in Table 1. Movement frequencies across the 4 types of movement in the two legs were highly symmetrical so all four movements occurred with similar frequencies ($F(3,84) = .630$, $p = .598$). The difference in overall average movement frequency between outcome groups (TD versus CP plus DD) was not statistically significant at 2 months CA ($t(13) = .029$, $p = .977$). The difference in overall average movement frequency between experimental and control groups was also not statistically significant at baseline ($t(3) = 1.125$, $p = .281$). Thus leg movement frequencies between groups were not significantly different at entry to the study.

Leg Movements While Tethered to the Mobile at Four Months CA

Table 1 and Table 2 show the mean frequencies of each leg movement and of the total of the four leg movements at 4 months CA. The difference in overall average movement frequency between outcome groups (TD versus CP plus DD) was not statistically significant at 4 months CA ($t(12) = 1.645$, $p = .126$). Although infants with CP had movement frequencies averaging 35.24 (SD 6.74) in the control group (N=2) and 61.87 (SD 55.73) movements/minute in the exercise group (N=2), the difference in overall average movement frequency between control and experimental group subjects was also not statistically significant ($t(12) = .149$, $p = .884$). Thus, no effect of exercise or of differences based on developmental outcome was discernible.

Longitudinal Analysis of Change With Age by Outcome and Experimental Group

Movement Frequency—The repeated measures ANOVA showed no differences in average total movement frequency across all four movements between age groups ($F(1,7) = .219$, $p = .654$), the 2-way interaction of age and experimental group ($F(1,7) = .984$, $p = .354$) and age by outcome ($F(2,7) = .714$, $p = .522$), or the 3-way interaction among age, experimental group, and outcome ($F(2,7) = 2.90$, $p = .121$). The mean difference between control and experimental group children with CP was −66.04 with a SE of 31.42 (95% confidence interval −140.34 to 8.26, $p = .074$) with the average movement frequency being higher in the infant with CP at 4 months CA following two months of tethered-kicking exercise than in control group children with CP.

Figure 3 shows the total of the four movement frequencies/minute of each child at 2 and 4 months CA plus the single data points of the 3 infants with CP who have missing data at one of the two data collection times. The graph shows that all infants with TD in the exercise group increased their kicking frequency between 2 and 4 months CA. All 3 children in the experimental group who had TD at 12 months CA were either walking independently or with just one hand held.[4] One child in the control group with a 2-month movement frequency of 48.3/minute was the most exuberant kicker at 4 months CA with a movement frequency of 118.3 movements/minute (Subject C1,Table 1). This child was an exceptionally high performer (75th percentile) on the AIMS at 12 months and the only child in the control group who was walking independently at 12 months CA.[4] The other 4 children with TD in the control group show smaller increases or decreases in movement frequency across the two ages, and none was walking at 12 months CA. Children with DD (E4 and C8, Table 1) show declining or stable movement frequencies across time. Unfortunately data points are missing for two children with CP at 4 months and one child with CP at 2 months CA. We do know that the latter (E6) with an average movement frequency of only 22 movements/minute at 4 months CA did not receive the expected dose of intervention because of a very challenging situation in the home. Nevertheless, it is apparent that the two children with CP in the control group showed a decline in overall movement frequency over time, in one case from 79.5 at 2 months CA to 40/minute at 4 months CA (Subject C7) and in the other (Subject C9) from 54.8 to 30.5/minute (mean difference 31.91, SE 17.88, p =. 117, 95% confidence interval −10.368 to 74.185) while one child with CP in the experimental group (E7) increased movement frequency from 57.2 movements/minute at 2 months CA to 101.3 movements/minute at 4 months CA. The correlation between movement frequency at baseline (2 months CA) and the TIMP score at 4 months CA was not statistically significant ($r(13) = -.006$, $p = .984$), but leg movement frequency at 4 months CA was significantly correlated with the TIMP score at the same age ($r(11) = .54$, $p = .047$). Thus children with better functional motor performance at 4 months CA had higher rates of leg movement.

Inter-limb Coordination Patterns—Figure 4 shows the change in proportion of movements that involve synchronous hip flexion of both legs across time and between experimental groups and developmental outcomes. All children with TD increase the proportion of kicks that are synchronous between 2 (mean 32%, SD 16%) and 4 months CA (mean 56%, SD 9%), as do the children with DD (20% to 32% and 35% to 39%, respectively). Children with CP in general show proportionally fewer synchronous kicks over time (mean 31%, SD 13% at 2 months compared to 19%, SD 15% at 4 months). Overall, infants with TD had a greater proportion of synchronous movements at 4 months CA when compared with infants whose outcome was CP or DD ($t(12) = 4.865$, $p < .0001$). The repeated measures ANOVA, however, showed no differences in the proportion of kicks that were synchronous between age groups ($F(1,7) = 1.477$, $p = .264$), the 2-way interactions of age and developmental outcome ($F(2,7) = 2.867$, $p = .123$) and age and experimental group assignment ($F(1,7) = .007$, $p = .936$), or the 3-way interaction of age, group and developmental outcome ($F(2,7) = .071$, $p = .932$). In pairwise comparisons of infants with TD the proportion of synchronous kicks increased over time in the control group (mean difference −.226, SE .088, $p = .037$, 95% confidence interval −.434 to −.018) and in the experimental group (mean difference −.260, SE .133, $p = .055$, 95% confidence interval −.529 to .008). Furthermore, in the experimental group, the child with CP did not differ from the children with TD in the proportion of kicks that were synchronous at four months (mean difference .179, SE .108, $p = .367$, 95% confidence interval −.158 to .516) while children with CP in the control group showed a lower proportion of synchronous kicks than the children with TD at four months (mean difference .369, SE .078, $p = .007$, 95% confidence interval .125 to .613). Thus children except for those in the control group with

CP tended to increase the proportion of kicks that were synchronous between 2 and 4 months CA but the overall differences between groups were not statistically significant.

Clinical Observations

Although the focus of the observations in this study was sagittal plane movements of the hip, i.e., hip flexion and extension, infants in the study engaged in a wide variety of movement types. In the videos at 4 months CA all infants with TD were observed to also make arm movements, including elegant wrist rotations and finger movement and grasping of the knees; one child reached up toward the mobile, and he and another child also lifted the legs and grasped the feet repeatedly. Several infants, both TD and CP, performed mouthing movements while kicking.

Support of the ankles by the sling tended to place the hips in relatively more abduction and external rotation at 4 months CA, and all infants with TD or DD were observed to rub their feet together frequently as well as perform knee extension movements of varying degrees from a 90-degree flexed-hip position. Five of the 8 infants with TD also performed bridging movements, i.e., pushing down with the feet and extending the hips off the support surface. Only one infant with CP ever rubbed the feet together so in most cases kicks in these children occurred relatively more in the sagittal plane. One child with CP (C7) was observed to extend the knee from a flexed hip only in the left leg. This child had a TIMP score of −2.81 and lacked fidgety movements at 4 months CA, and was classified as GMFCS level II or mildly involved (sitting alone and standing with support) at 12 months CA. No other infants with CP showed knee extension from a flexed-hip position while tethered to the mobile and none showed bridging with the legs. Of note, however, is that movement of the children with CP was [u]not yet characterized by hip adduction and internal rotation while supported in the slings. Lack of variety was more notable than abnormal patterns of coordination, but while 6 of 8 children with a typical developmental outcome showed fidgety general movements on the TIMP at 4 months CA, only 2 of 8 with an outcome at 12 months of CP or DD showed fidgety movements at 4 months CA.

Movement variety was less evident in all children at 2 months CA. Half of the children showed foot rubbing, but only children with TD and one child with DD showed knee extension from a flexed-hip position.

Discussion

Task-oriented Training

Therapy for children at risk for CP should be directed at muscle activation, development of sensory and motor control processes, and motor learning to encourage optimal growth and development while minimizing maladaptive changes to the neuromusculoskeletal system.[18] We are not aware of other published research on children with PBI aimed at facilitating leg movements during a functional activity in early infancy, but similar tasks are under investigation in children born term or preterm. For example, Heathcock and colleagues[19] have shown that foot reaching can be learned by preterm infants.

A second approach is that of Fetters consisting of behavioral shaping to decouple joint movements such that knee extension with hip flexion is facilitated.[10] The ability to decouple knee movements from hip movements emerges as infants exploit passive dynamics in coordination of hip and knee motions.[20] Decoupling has been successfully shaped in a laboratory-based intervention in children born preterm or term, and the group is currently developing a system for home use with children with disabilities.[10] Our anecdotal observations showed that children with TD spontaneously produced knee extension from a flexed hip, especially at 4 months CA, but this pattern of coordination was observed only once at 4 months CA in a child with mild CP involvement and not at all in other subjects with CP. Thus training should begin early.

Feasibility of Tethered-kicking Exercise Training in Children with PBI

Because our analyses of the effects of the intervention on overall motor performance did not show statistically significant differences between groups,[4] we undertook this behavioral analysis to assess whether children had the ability to attend to the task and use leg movements to make the mobile move. Results support the feasibility of the intervention. With the exception of one infant at 2 months CA, children remained alert and attentive to the task for several minutes. Children with DD or CP did have more difficulty with maintaining a midline head position than infants with TD but nonetheless met the criterion for having achieved midline head control as defined by Dusing and colleagues.[21] Thus attention and head control do not seem to be limiting factors in the performance of infants with PBI. In fact, we conjecture that attending to the mobile helps children with CP to maintain a midline-head position.

With both legs supported by the tethers, infants were able to explore a wide variety of movement options. For example, alternate kicks can make the mobile twirl while synchronous hip movements make the toys hanging from the mobile bounce. Infants kicked, creating mobile movement, albeit with varying frequencies. Higher movement frequency was significantly correlated with functional performance on the TIMP at 4 months CA.

Patterns of Inter-limb Coordination

In a study of spontaneous leg activity in infants born at term or preterm with very low birthweight, Jeng and colleagues reported a decline in the proportion of kicking movements that were alternate between 2 and 4 months of age.[7] Our work demonstrates a similar finding in a functional movement activity. Our results for children with the outcome of CP or DD are, however, different from those reported by Jeng and colleagues. They reported that children with delayed development of walking (median age of 14 months CA) tended to have more synchronous kicking than children with TD as well as a high kick frequency at 4 months CA. In our study children with TD showed a greater proportion of task-related kicks that were synchronous at 4 months CA as well as a highly variable but generally larger movement frequency when compared to children with CP or DD. The child with CP in the exercise group, however, did not differ from children with TD in the proportion of kicks that were synchronous while children with CP in the control group differed from those with TD. These differences in findings may result from differences in the task: spontaneous kicking versus movements in a rewarding functional activity. The positioning and measurement protocol also was different: 20 seconds of spontaneous activity selected for when the most kicking occurred, head fixed in midline, in Jeng and colleagues' work,[7] while we supported the infants' legs to lower the influence of the force of gravity, allowed free head movement, and assessed the entire period of awake behavior.

Efficacy of the Intervention

In this small sample, results comparing intervention groups across age and outcome at 12 months did not reach statistical significance, but we believe that the findings reported here along with the potential differences in age of onset of locomotion between exercise and control groups[4] warrant further examination. In other reports we have shown that we can successfully engage the families of these children with PBI in a year-long longitudinal study of intervention and assessment, including measurement of maturation of white matter in the brain at 12 months CA using diffusion tensor imaging as a potential assessment of brain plasticity in response to intervention.[4,5]

Limitations of the Study

In addition to the problem of missing data, the sample for this study was small and the outcomes heterogeneous. We used a single camera to film the infants in homes with varying quality of lighting, ambient noise and activity. The analysis of movement employed was based on observed behavior rather than a formal kinematic assessment. Learning was not assessed so we do not know the extent to which cognitive issues affected performance. As previously reported, the families in this study did not always carry out the intervention at the requested frequency and intensity, so the dosage received was typically only about twice a week for 5 minutes according to parent diaries.[4] Other physical therapy did not generally occur as early as expected and, in half of the infants, did not occur at all in the first year.[4]

Plasticity in Adaptation to Brain Injury

Our aim is to speed the recovery of infants with the potential for adaptation to PBI with typical motor development and to reduce the severity of CP or DD in those with permanent impairment of neural function. Whether our intervention begins early enough to change the course of motor development is a major question. Corticospinal tracts are already beginning to be myelinated by term age in the newborn and connections to spinal motoneuron centers allow coordination of leg movements from an early age.[22,23] Spontaneous leg movements have already been shown to be atypical by one month CA in infants with white matter damage,[6,8] and, whatever their pattern of coordination, the movements available will be used by the infant to drive development of activity-dependent areas of the cerebral cortex which may lead to secondary alterations of brain structure and function as well as muscle architecture and neural connections.[22,23] Except in those with severe brain damage, however, early movement differences can be subtle and highly variable;[3] when fidgety general movements fail to develop at 3–4 months CA, risk for CP can first be predicted with high sensitivity.[24] At 4 months CA all infants in this study with CP or DD at 12 months CA were also delayed in functional motor performance as measured with the TIMP and tended to show lower frequencies of leg movements while tethered to the mobile than infants with TD. Thus intervention beginning only at 3–4 months CA is possibly already far too late to alter the course of abnormal development of the musculoskeletal and neurologic systems in babies with permanent brain damage.

Conclusions

The results of this work and other previously reported aspects of the study[4,5] demonstrate that the home-based, task-oriented intervention approach using commercially available toys we designed for infants with PBI is feasible to implement from 2–4 months CA. A majority of the children in the study increased the frequency of leg movements between 2 and 4 months CA in response to being tethered to the mobile while supported in an infant bath seat. Further research is necessary to refine the protocol, increase compliance with the exercise, and evaluate the efficacy of the intervention.

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Fig. 1.
The Exercise. The mobile (sound and movement deactivated) is attached to a wooden stand that slides under the infant bath seat in which the child is seated. Tethers are attached to each of the infant's ankles with adjustable Velcro straps. A noise-making toy hangs from the mobile to provide both visual and auditory feedback when the infant kicks.

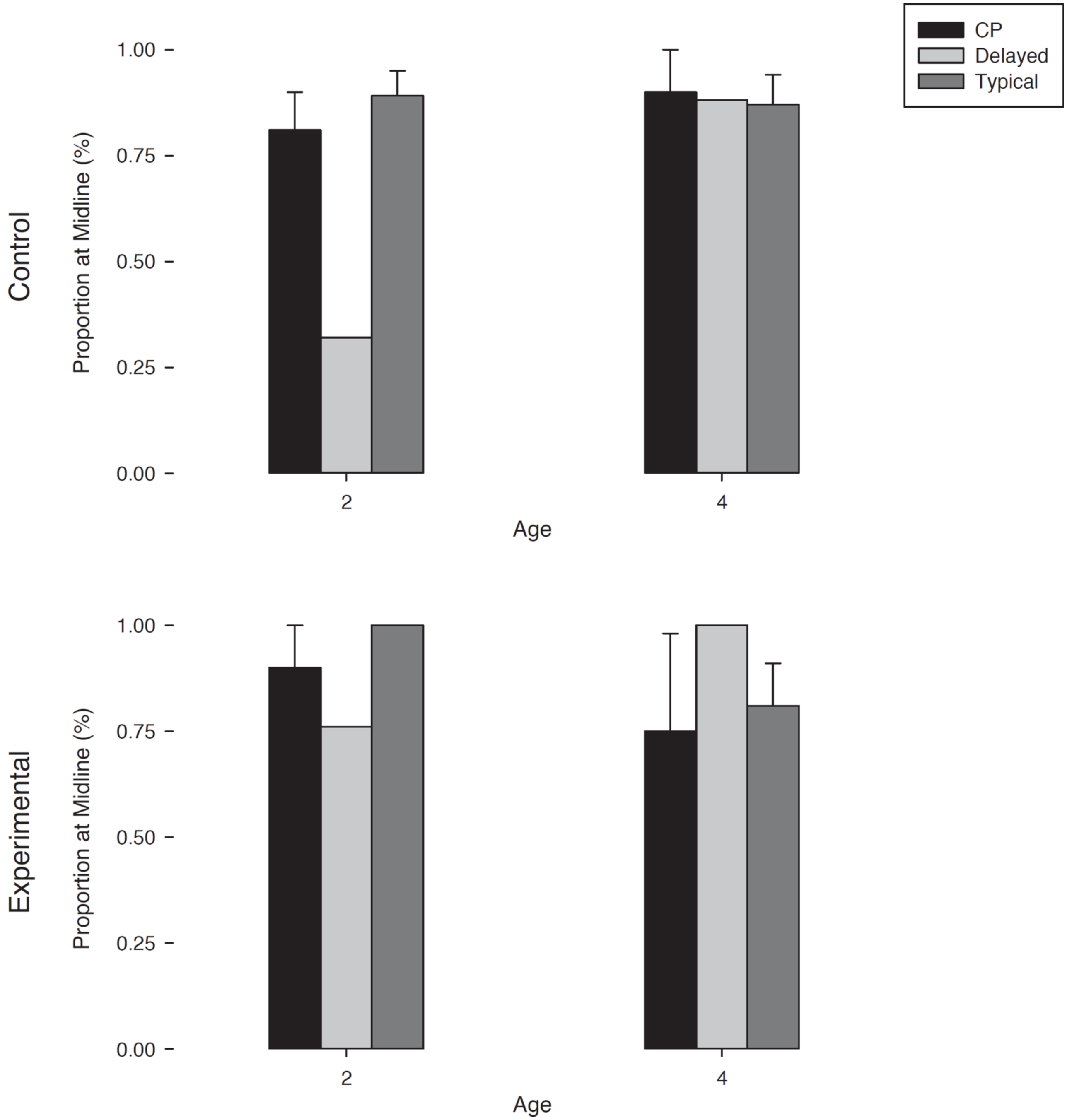

Fig. 2.

Average Proportion of Time With Head in Midline by Age, Outcome, and Group Assignment.

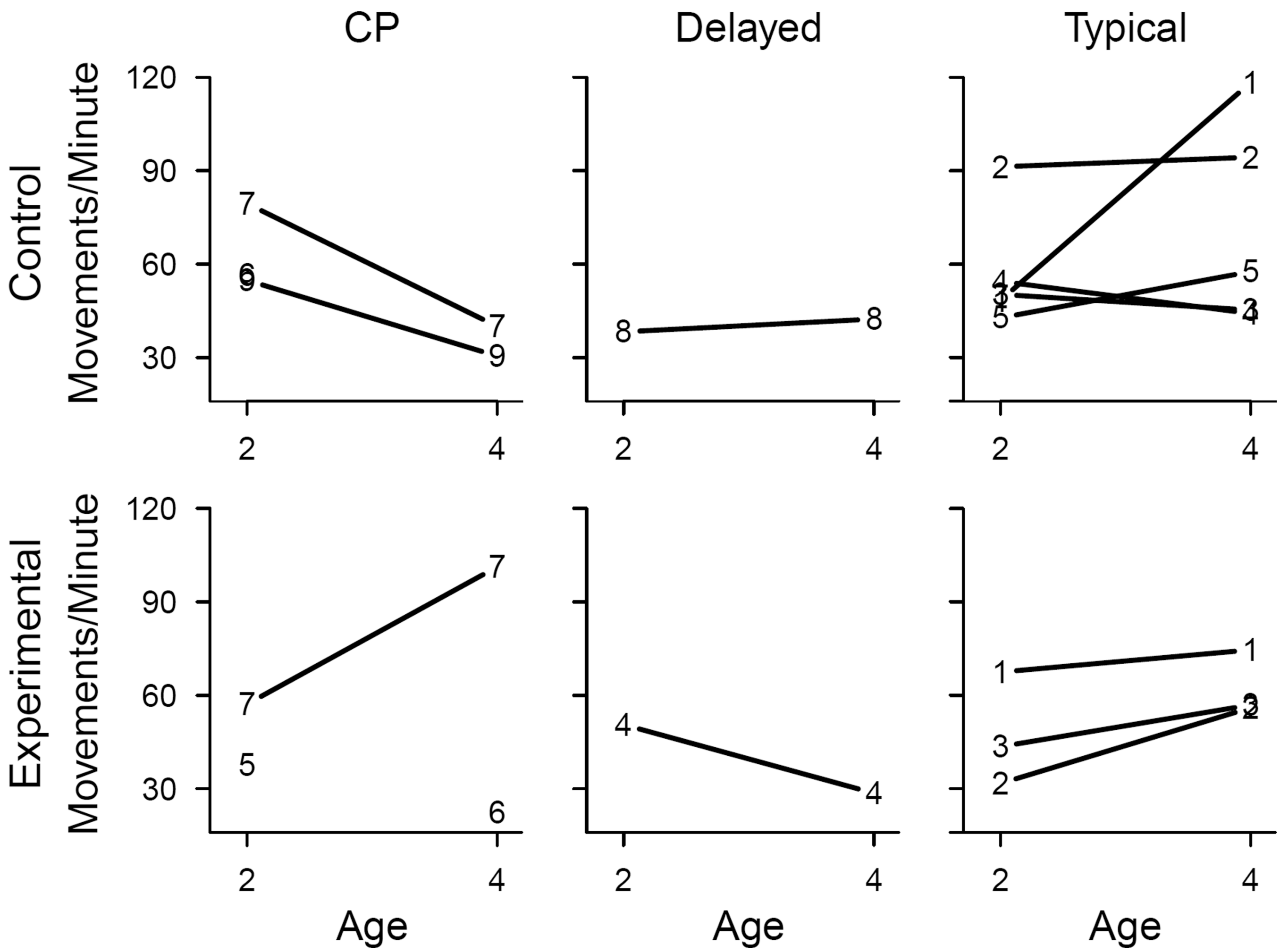

Fig. 3.

Longitudinal Change in Total Leg Movement Frequency of Individual Children by Age, Outcome, and Group Assignment. CP, cerebral palsy. Numbers in the graph correspond with subject numbers in Table 1.

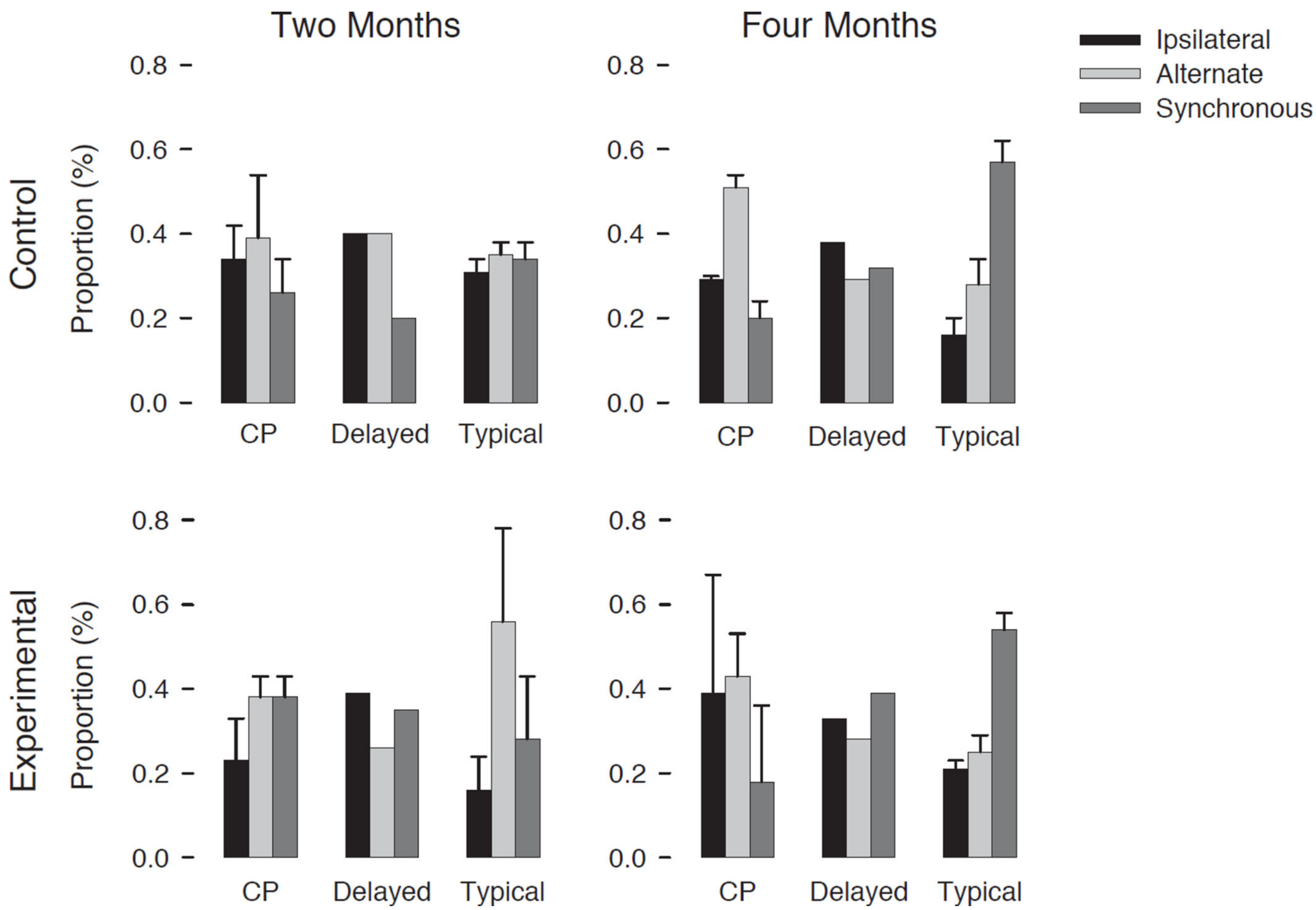

Fig. 4.
Inter-limb Coordination Patterns by Age, Outcome, and Group Assignment: Proportions of Right Hip Flexion Movements That Were Ipsilateral, Alternate and Synchronous. CP, cerebral palsy.

Table 1

Description of Subjects Arranged in Order of AIMS Outcome at 12 Months CA and Experimental Group Assignment

Subject Number & Group	**E1**	**E2**	**E3**	**E4**	**E5**	**E6**	**E7**	**C1**	**C2**	**C3**	**C4**	**C5**	**C6**	**C7**	**C8**	**C9**
GA at birth	30	27	23	27	24	26	24	26	32	29	31	28	26	32	25	24
4 mo TIMP Z score	0.50	0.75	−0.56	−1.75	−2.75	−3.63	−1.06	0.06	−0.25	0.25	−0.69	−3.19	−1.74	−2.81	−1.00	−3.81
Movements /minute at 2 mo	67.5	31.7	43.5	50.6	37.6	------	57.2	48.3	91.2	50.3	54.5	42.8	56.4	79.5	38.3	54.8
Movements /minute at 4 mo	74.6	56.0	56.9	28.6	------	22.5	101.3	118.3	94.3	45.3	44.2	57.6	------	40.0	42.4	30.5
12 mo AIMS Z Score	0.80	0.53	0.24	−1.68	−3.42	−4.96	−5.39	0.53	0.24	0.10	0.10	−0.35	−3.70	−3.80	−5.09 (16 mo)	−10.10
Highest motor skill at 12 mo CA	Walks alone	Walks alone	Walks with one hand held	Creeps, takes steps with trunk support	Sits, stand with support	Rolls, does not sit alone	Does not roll or sit alone	Walks alone	Creeps, cruises	Creeps, takes steps with trunk support	Cruises, takes steps with trunk support	Creeps, cruises	Rolls, sits alone	Sits, stand with support	Walks at 16 mo	Does not roll or sit alone
GMFCS	---	---	---	DD	II	IV	V	---	---	---	---	---	II	II	DD	V

E, exercise group; C, control group; GA, gestational age in weeks; TIMP, Test of Infant Motor Performance; CA, corrected age; AIMS, Alberta Infant Motor Scale; GMFCS, Gross Motor Function Classification System; DD, developmental delay, no cerebral palsy

Table 2

Mean frequencies (movements/minute) for each of the four movement types and mean total movement frequencies by outcome and age

	2 months			4 months		
	N	Mean	*SD*	*N*	Mean	*SD*
Cerebral palsy						
Right Leg Flexion	5	15.95	9.05	4	9.54	3.72
Right Leg Extension	5	14.90	5.46	4	14.36	14.74
Left Leg Flexion	5	11.74	4.61	4	12.14	12.92
Left Leg Extension	5	14.51	4.65	4	12.51	7.68
Overall	5	57.10	14.87	4	48.55	35.87
Delayed						
Right Leg Flexion	2	10.36	4.38	2	9.88	3.19
Right Leg Extension	2	11.17	3.75	2	7.51	5.78
Left Leg Flexion	2	11.20	.41	2	10.36	1.41
Left Leg Extension	2	11.70	.18	2	7.72	2.18
Overall	2	44.43	8.70	2	35.48	9.74
Typical						
Right Leg Flexion	8	11.12	6.44	8	16.96	7.68
Right Leg Extension	8	14.29	6.18	8	15.15	6.20
Left Leg Flexion	8	13.99	5.97	8	21.30	8.19
Left Leg Extension	8	14.32	5.67	8	14.99	8.03
Overall	8	53.73	18.31	8	68.40	25.99