Early Spontaneous Leg Movements in Infants Born with and Without Myelomeningocele

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Purpose: To compare quantity and quality of spontaneous leg movements during early infancy in babies with myelomeningocele (MMC) and babies with typical development (TD). **Methods:** Nine infants with MMC and 12 with TD moved spontaneously while supine for 5 minutes at ages 1, 3, and 6 months. We used a 6-camera system to monitor leg movements. Resultant leg displacement and velocity were used to determine movement frequency and, for each movement, duration, distance, peak velocity, jerk, and number of acceleration peaks. **Results:** Movements of infants with MMC were shorter in duration with fewer acceleration peaks than their peers. Asymmetrical interlimb frequencies in infants with TD resulted in one leg moving more than the other, which was similar to the lower symmetrical interlimb frequencies of infants with MMC. **Conclusions:** Infants with MMC show depressed movement activity. Further research is needed to determine if therapy can facilitate spontaneous activity and leg control. **(Pediatr Phys Ther 2008;20:137–145)** *Key words: child development/physiology, human movement system, infant/physiology, kinesiology/applied, leg/physiopathology, motor activity/physiology, movement/physiology, time factors*

INTRODUCTION

Advances in women's preconception healthcare and nutrition, especially folic acid supplementation, have succeeded in reducing the number of babies born with myelomeningocele (MMC), particularly in economically advanced nations. Nevertheless, this remains the most common neural tube defect and the incidence of babies born with MMC ranges in the world today, from 2.92 per 1000 births in Northern China[1] to 0.20 per 1000 births in the United States.[2] The lesion levels at which MMC occurs most frequently are lumbar and sacral, making development of lower trunk and leg control particularly difficult, if not impossible, to accomplish. In a large sample studied by Williams et al[3] approximately 56% of children with L1-L3 level lesions achieved walking but only 32% remained walkers at 10 years of age. Eighty-four percent of those with L4-L5 levels walked but only 73% of these retained this function after age 9 years. The outcome for sacral level involvement is much better, nearly 100% achieve and retain walking. On average, if walking is achieved, onset is delayed by about 2 years, compared with infants with typical development (TD), and often involves the use of leg braces or other assistive devices.[4,5] Walking is, typically, unstable and energy costly.[6,7]

By birth, and often before birth, we know if MMC has occurred. In the US surgical repairs routinely occur within 24 hours of birth or even prenatally.[8] But surgery repositions the spinal cord in the vertebral column and protects it; it does not fix the damage already done to the developing nervous system. Early nervous system development and organization are strongly influenced by infants' own attempts to move and explore their environments.[9,10] During repeated cycles of perceiving and acting, young infants discover how to control and adapt their movements, strengthen connections between populations of sensory and motor neurons, and build muscle strength.[11] When the neural substrates of this process are compromised, as is the case for infants born with MMC, the challenge of optimizing development of the neuromotor system and learning to function becomes much more difficult.

Numerous researchers have examined the spontaneous leg movements of healthy infants and described, in detail, the changes in behavior over time.[12–14] Of special

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interest are shifts in performance that occur as infants grow older and differences between the limb movements of healthy and disabled populations, which provide insights to the processes underlying and affecting the acquisition of neuromotor control.[15,16] For infants with MMC who experience delays and difficulties acquiring leg control and locomotion, research in this area is less extensive but some information is available. For example, 16- to 24-week-old fetuses with MMC show leg movements as frequent as their peers without MMC.[17,18] Sival et al[19] report that within a few days after birth neonates with MMC show a reduction in leg movements and fewer movements than neonates with TD. Chapman[20] studied 4- to 8-month-old infants with and without MMC and found an overall group difference in frequency of leg movements, but not movement velocity or distance. The existing data suggest a nonlinear developmental course with unique continuities and discontinuities but one that has glaring gaps as well. We know little about the quality and quantity of spontaneous leg movements of infants with MMC over the first few months of postnatal life and no one has examined the differences between their legs. However, we do know that damage to the spinal cord is not likely to uniformly affect the neuromuscular system in the medial-lateral and anterior-posterior regions.[21] Yet, an important aspect of learning to control one's limbs is discovering how to differentiate and adapt interlimb movements to fit particular contexts and tasks. Thus it is important to examine each leg and the development of interlimb control.

Therefore, to fill in gaps in the developmental profile of infants born with MMC and to lay the groundwork for further experimental studies and tests of the effect of therapeutic interventions we designed a longitudinal study of their early spontaneous leg movements. Our goal was to precisely monitor the quantity and quality of spontaneous leg movement in infants with MMC and with TD, when supine, over the first few months after birth. Further, we distinguished the movements of each leg to address movement symmetry.

METHOD

Participants

In a large longitudinal study conducted over the course of 3 years, 12 infants with MMC and 12 with TD participated in our study of upright stepping and supine spontaneous leg movements. The spontaneous movement portion of this study, the data we present here, represent a descriptive comparison of 2 groups, 9 infants with MMC and 12 with TD. Two infants with MMC enrolled in our larger study before spontaneous movement was included as a portion of data collections. In addition, all of the spontaneous movement data from 1 participant in the MMC group and several individual trials from the dataset of infants with MMC and TD were lost because of technical problems. Only infants with MMC who had no known chromosomal or CNS abnormalities with the exception of the anomalies known to be associated with MMC, including hydrocephalus and Arnold-Chiari II malformation, were included. Lesion levels were limited to lumbar and sacral; gestational ages at birth were 36 or more weeks for all but 1 infant, who was born at 34 weeks and entered the study at 1 month corrected age. Infants with TD were without known cognitive, sensory, or motor disabilities. During their involvement in the study presented here, 3 infants received no physical therapy, three received support services via the Michigan Department of Education's Early On program approximately once per week starting between the 3rd and 6th month, and 3 received services from a PT once per week starting on or after 3 months of age. Table 1 contains mean and individual characteristics of infants in both groups including the structural level at which MMC occurred, occurrence of hydrocephalus, shunting, and club foot among infants with MMC, the leg preference of each infant with MMC, gestational ages at birth, birth weights, and birth lengths.

Infants with TD were recruited through newspaper advertisements, fliers, and word-of-mouth and lived in the

TABLE 1
Infant Characteristics

Subject		Hydrocephalus	Shunted	Arnold Chiari	Club Feet	More Affected Leg	Gestation Age (wk)*	Birth Weight (kg)	Birth Length (cm)
	Structural Level								
1	L1/L2	Y		Y		L	37	3.52	53.34
2	L2/L3	Y				L	34	2.10	44.45
3	L3	Y	Y	Y	Y	L	38	3.06	47.00
4	L3	Y	Y			NA	40	3.81	54.61
5	L4/L5			Y		R	38	3.27	45.72
6	L5/S1					NA	37.5	3.52	55.88
7	L5/S1	Y	Y			NA	36.5	2.95	48.90
8	S1					NA	36.5	3.71	49.53
9	S1					L	37	2.83	48.26
	Myelomeningocele group	Mean					37.17	3.20	49.74
		SD					1.60	0.53	4.02
	Typical development group	Mean					39.21	3.58	52.30
		SD					0.81	0.44	2.58

* Statistical significance.

Ann Arbor, Michigan area. Infants with MMC were referred by physicians at the University of Michigan hospital and St. Vincent Mercy Children's Hospital of Toledo, Ohio and lived within a 3-hour radius of our laboratory. Visits to our lab coincided, for infants with MMC, generally, with visits to their primary care physicians for checkups. Approval for the study was granted through the Institutional Review Board at the University of Michigan and parents provided informed consent before their infants' participation.

Procedure

Data presented here are part of a larger study for which babies visited our lab for testing at 1, 3, 6, 9, and 12 months of age, at walking onset, and after 3 months of walking experience. For the purposes of the issues on which we focus this article, that is, spontaneous leg movements in supine, we collected data only up to the age when infants began to roll over. For all but 2 infants with TD this occurred by 6 months of age. All test sessions in the larger study included 2 contexts: upright (U)—in which we attempted to elicit newborn steps and treadmill steps, and supine (S)—in which we monitored spontaneous leg movements as infants lay supine on a firm surface. Infants experienced context U first, in a session lasting about 40 minutes, including approximately 5 minutes (total) of measured activity. We administered context S next, which required about 25 minutes, including 5 minutes of measured activity. In context U infants wore 4 electromyographic electrodes on one leg and 10 reflective markers distributed across both legs. In context S infants wore only 6 reflective markers.

When infants arrived at our lab we removed their clothing and diaper and prepared them for testing in the first context, U. Following all U testing we removed the EMG electrodes and repositioned the reflective markers, placing them on the lateral surface of the greater trochanter, ventral surface of the patella, and ventral surface of the third metatarsal. We found via pilot work that these marker locations provided optimal camera views of the legs when infants were supine. We placed infants on their backs on a towel-covered firm surface. Before the trial began infants' heads were positioned in midline, then released; we held their legs extended and parallel for 10 seconds at the start of each trial before releasing them. Parents and researchers maintained conversation and music played in the background but no direct interactions with infants were allowed unless the infant became quite upset, at which point attempts were made to calm the infant via verbal (only) communication. Three trials of 2-, 2-, and 1-minute duration were collected. Individual trials were limited in duration because of computer storage issues and to allow 'breaks' for infants.

All kinematic data were captured by a 6-camera Peak Motus (Peak Performance Technologies Inc., Centennial, CO) system sampling at 60 Hz with 3 cameras positioned on each side of the movement space and 1 digital camera positioned to record the entire session. All cameras were synchronized and we calibrated the space before each session; the summed error measurement for calibrations was limited to ≤ 0.008 m.

Data Reduction

Ultimately, we were able to analyze 85% of trials collected for infants with MMC and 93% for those with TD; loss was due to technical difficulties with cameras, positions of the limb that were not visible, or noise in the data. We converted the analog signals captured by Peak cameras to 3-dimensional digital data via Peak Motus software. Digital coordinates were reviewed to eliminate tracking errors and reflection errors. For individual joint markers 3-dimensional displacement data and linear velocity data were filtered with a second order Butterworth filter (6 Hz). We used a custom-written MATLAB (Mathworks, Natick, MA) program to interpolate small gaps in continuous data. Because we believe that spontaneous movements are better represented in all 3 axes, rather than in 1 plane at a time, we converted our data to resultant values. Three-dimensional linear velocities were converted to resultant velocity (cm/s) and 3-dimensional displacement coordinates to resultant displacements with the following formulas:

$$\text{Resultant velocity} = \sqrt{\dot{x}^2 + \dot{y}^2 + \dot{z}^2}$$

$$\text{Resultant displacement} = \sqrt{x^2 + y^2 + z^2}$$

where $\dot{x}$, $\dot{y}$, and $\dot{z}$ are velocities in the anterior-posterior, medial-lateral, and vertical directions and x, y, and z are 3-dimensional displacements in the anterior-posterior, medial-lateral, and vertical directions, respectively. We then entered the resultant velocities for the foot marker into a custom-written MATLAB program to extract events that qualified as foot/leg movements from nonmovements. We defined a movement as occurring when velocity reached at least 15 cm/sec for 0.10 seconds. In addition, if the resultant velocity exceeded 15 cm/sec, lasted 0.10 seconds, and superimposed on that movement another velocity peak occurred, that peak had to exceed 4 cm/sec to be considered another discrete movement. Figure 1 illustrates a time series with resultant velocity peaks highlighted.

After identifying all movements that met our criteria we calculated 6 movement variables: frequency (number of movements produced per second), and for each movement we calculated duration, distance, peak velocity, jerk, and number of acceleration peaks. The first 3 variables were included to reflect overall efforts to initiate movement (activity level) and vigor. The second 3 variables were included to address movement smoothness or control. We distinguished the more and less affected legs of infants with MMC, based on parental reports in addition to surgical notes, including references to things such as joint abnormalities or limb weaknesses. Because we had no reason to assume underlying neurological asymmetries for infants with TD we arbitrarily selected the right leg of infants with TD to be paired with the less affected leg for infants with MMC. For some infants with MMC there was no support for one leg being more affected

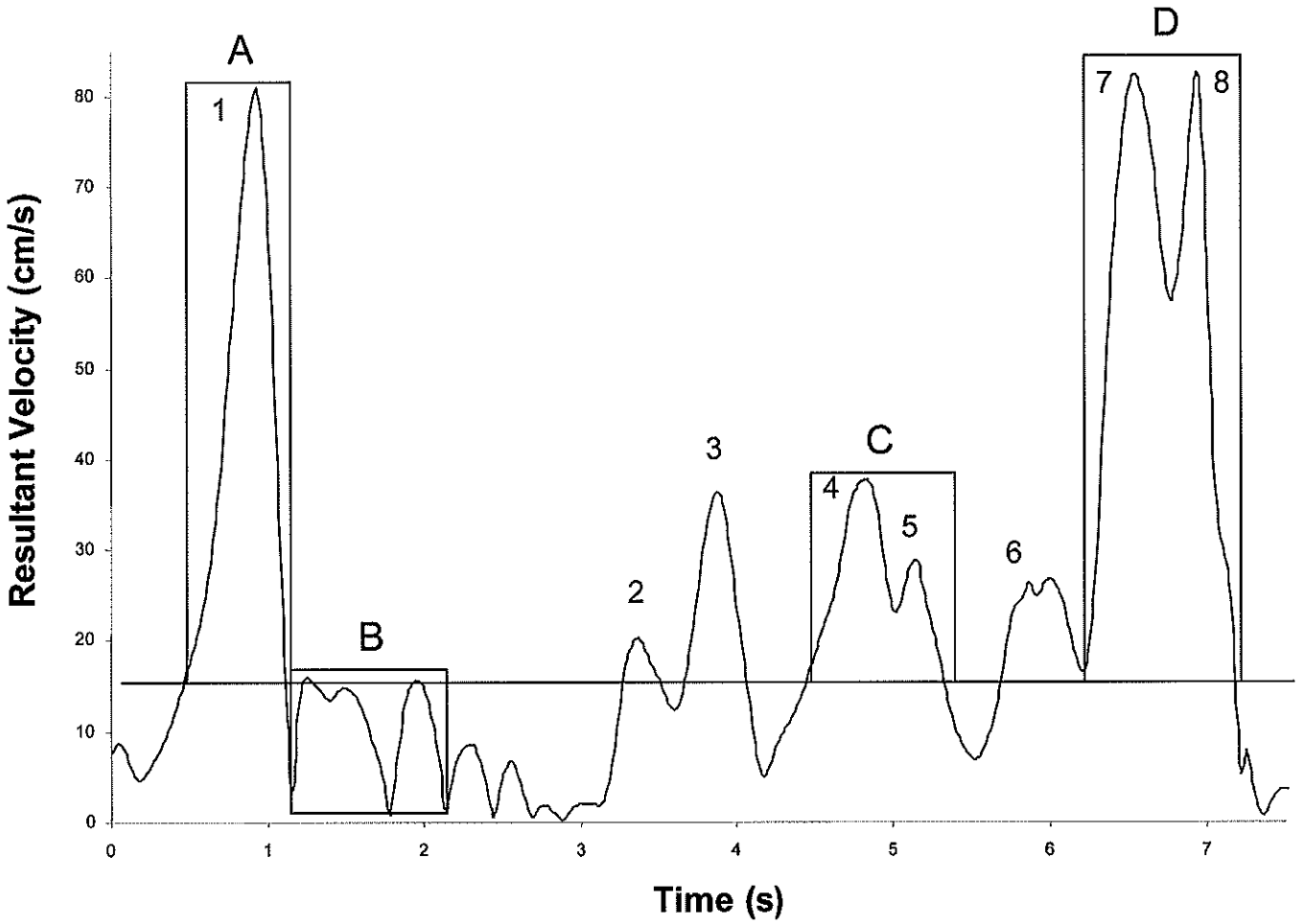

Fig. 1. Seven and a half seconds of exemplar resultant velocity data that includes 8 movements, with each movement identified by number for the reader. A shows resultant velocity >15 cm/sec for >0.10 seconds; B, resultant velocity >15 cm/sec for <0.10 seconds, and thus, not a movement; C and D, resultant velocities >15 cm/sec for >0.10 seconds with directional changes >4 cm/sec, creating 2 distinct movements (4, 5 and 7, 8).

than the other we applied the same approach as with infants with TD, that is, the right leg was considered less affected. Distribution of data by infant is provided in Table 1.

Statistical Analysis

We used a 2 (group) × 2 (leg) × 3 (age) ANOVA (SAS, Cary, NC) with repeated measures on the third factor to determine statistical significance, set at 0.05, for all 6 dependent variables (frequency, duration, distance, peak velocity, jerk, and number of acceleration peaks). In the following section statistics only for those results that were statistically significant are included.

RESULTS

Our results are organized in 2 sections by order of increasing depth of analysis. First we focus on the more conventional simple descriptives of the movements made (frequency, duration, and distance). Second we address velocity, jerk, and acceleration characteristics.

Movement Frequency, Duration, and Distance

Figure 2 shows that overall mean frequencies for infants with TD and MMC were 0.87 per second and 0.51 per second, respectively. However, variability within groups was also high and no main effects for frequency reached statistical significance. The group × leg interaction effect was significant, $F(1,19) = 6.58, p < 0.02$. Figure 2 suggests that the frequency with which infants moved each leg varied by group. Infants with MMC moved both legs at similar but lower frequency levels across age while, across this period of time their peers with TD moved one leg more than the other.

We considered the possibility that movement frequency was confounded by size of movement. To investigate this we further categorized each movement made by its size (displacement), small, medium, or large. The distribution pattern within each of these groups was similar to that for the others. That is, the difference in frequency or lack thereof was not due to infants with MMC performing only small movements and those with TD performing a particularly high proportion of large movements.

Our analysis of the mean duration of movements infants produced resulted in a significant main effect for group [$F(1,19) = 6.47$, $p < 0.02$] and age [$F(2,16) = 6.79$, $p < 0.007$]. Figure 3 illustrates that the average movement duration for infants with TD was longer than

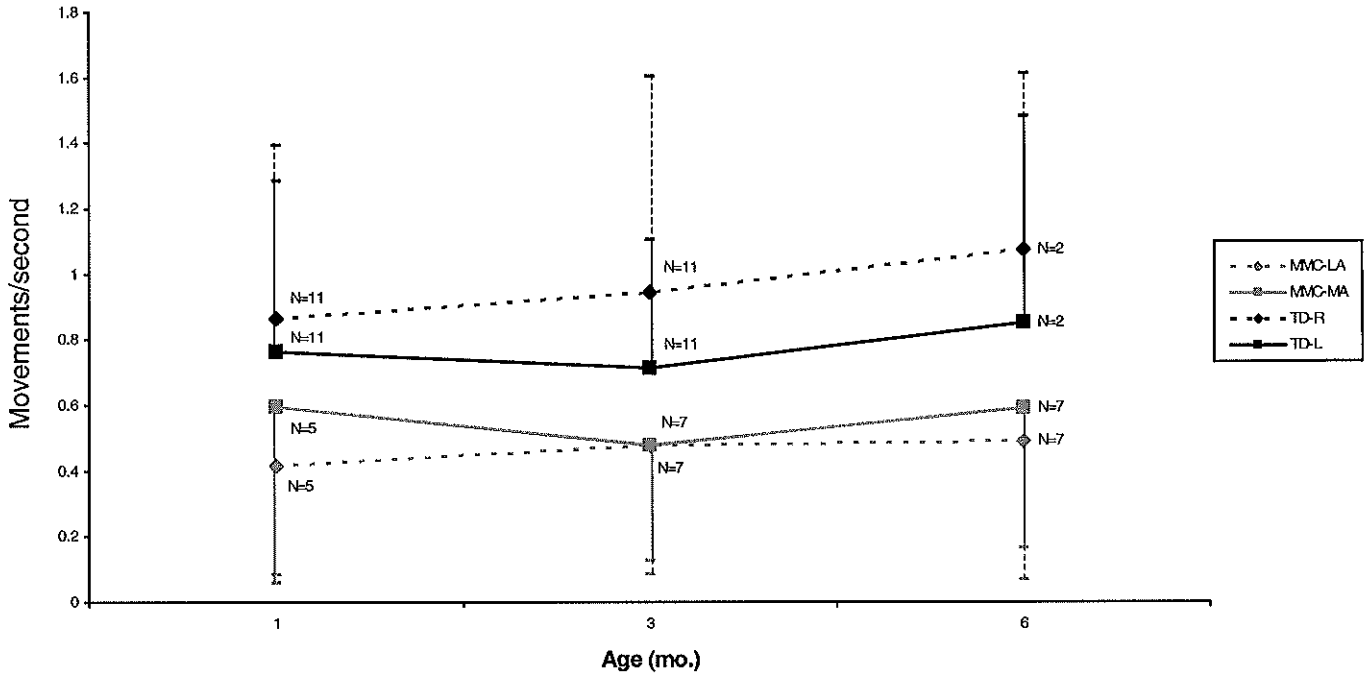

Fig. 2. Frequency of movement of the right and left legs of infants with typical development (TD) and the more affected (MA) and less affected (LA) legs of infants with myelomeningocele (MMC).

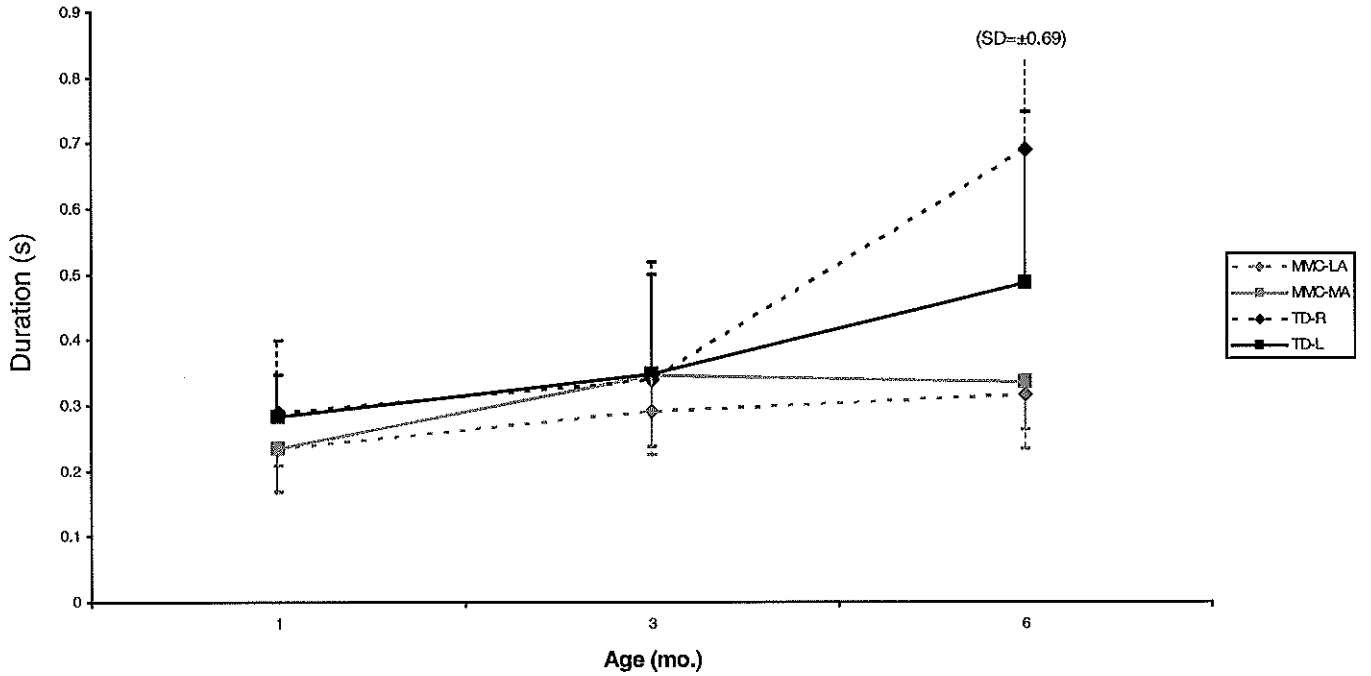

Fig. 3. Duration of movements produced by the right and left legs of infants with typical development (TD) and the more (MA) and less affected (LA) legs of infants with myelomeningocele (MMC).

that produced by infants with MMC. Further, both groups produced movements of longer duration as they grew older.

One effect was significant for the analysis of distance traveled, the leg main effect [$F(1,19) = 4.63$, $p < 0.04$]. Figure 4 shows that, on this factor both groups showed asymmetry, that is, in how far they displaced their leg with each movement, and they did so consistently across age. Infants with TD moved the R leg farther than their L leg and infants with MMC moved their less affected leg farther than their more affected leg.

Velocity, Jerk, and Acceleration

None of the main effects or interactions for peak velocity reached statistical significance.

For our analysis for jerk, or the smoothness of a movement, we obtained a significant leg main effect [$F(1,19) = 5.00$, $p <$

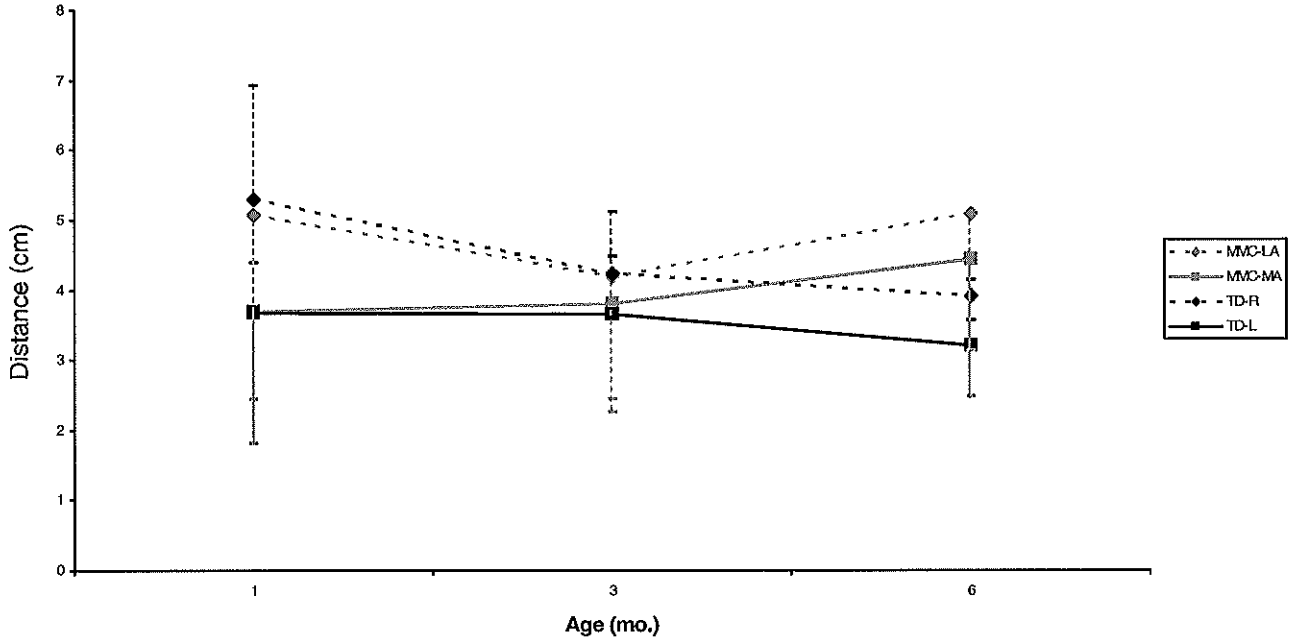

Fig. 4. Distance moved by the right and left legs of infants with typical development (TD) and the more (MA) and less affected (LA) legs of infants with myelomeningocele (MMC).

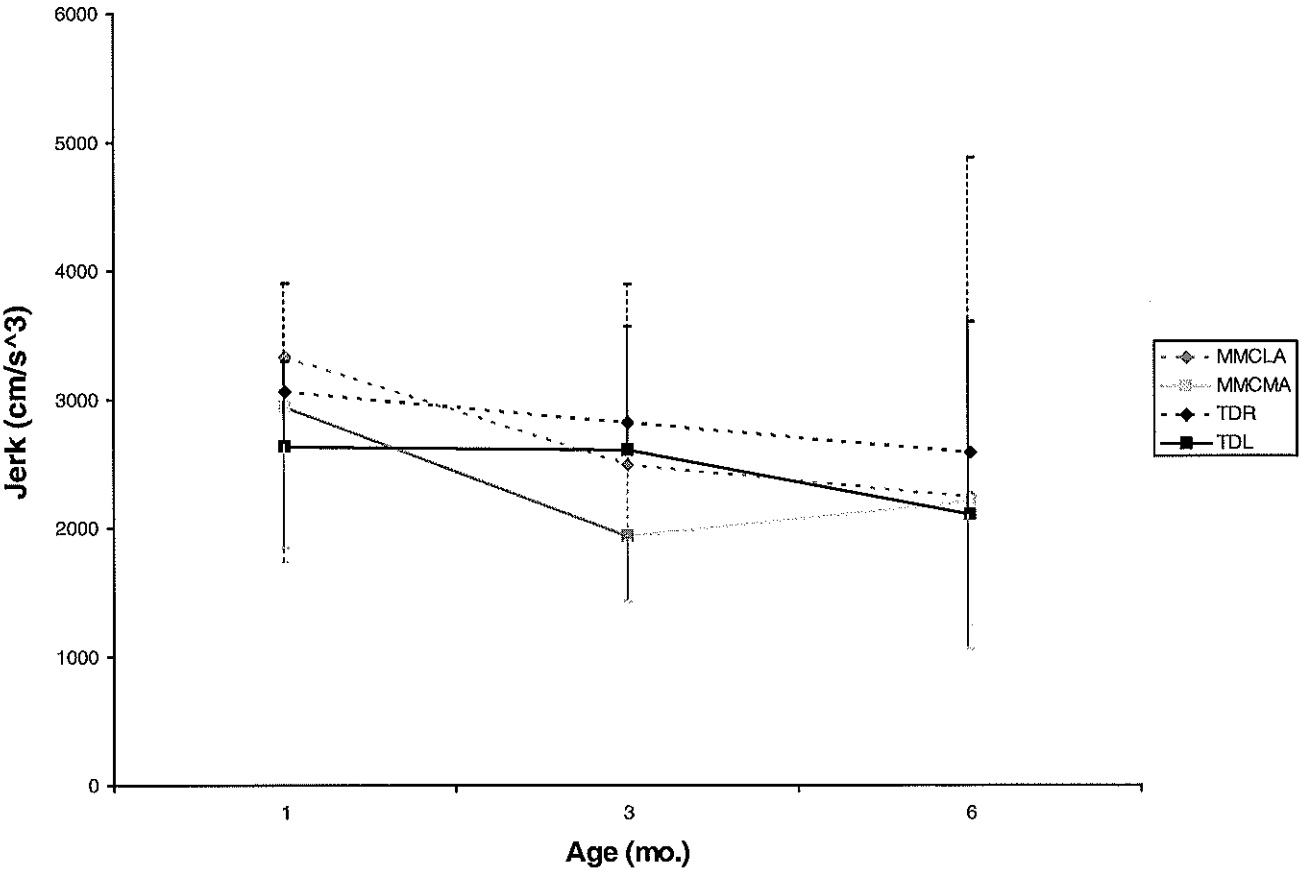

Fig. 5. Movement jerk produced by the right and left legs of infants with typical development (TD) and the more affected (MA) and less affected (LA) legs of infants with myelomeningocele (MMC).

0.04]. As we found for distance traveled, both groups showed asymmetry in the smoothness of their movements. Figure 5 demonstrates that infants with TD showed higher jerk values for their R leg than their L leg and for infants with MMC, higher jerk values were produced with their less affected leg than their more affected leg. Unlike the distance variable, we observed a trend toward a reduction in jerk with age [$F(2,16) = 3.06, p < 0.075$].

The outcome of our analysis of mean number of acceleration peaks per movement resulted in a significant group main effect [$F(1,19) = 7.65, p < 0.01$] and no other

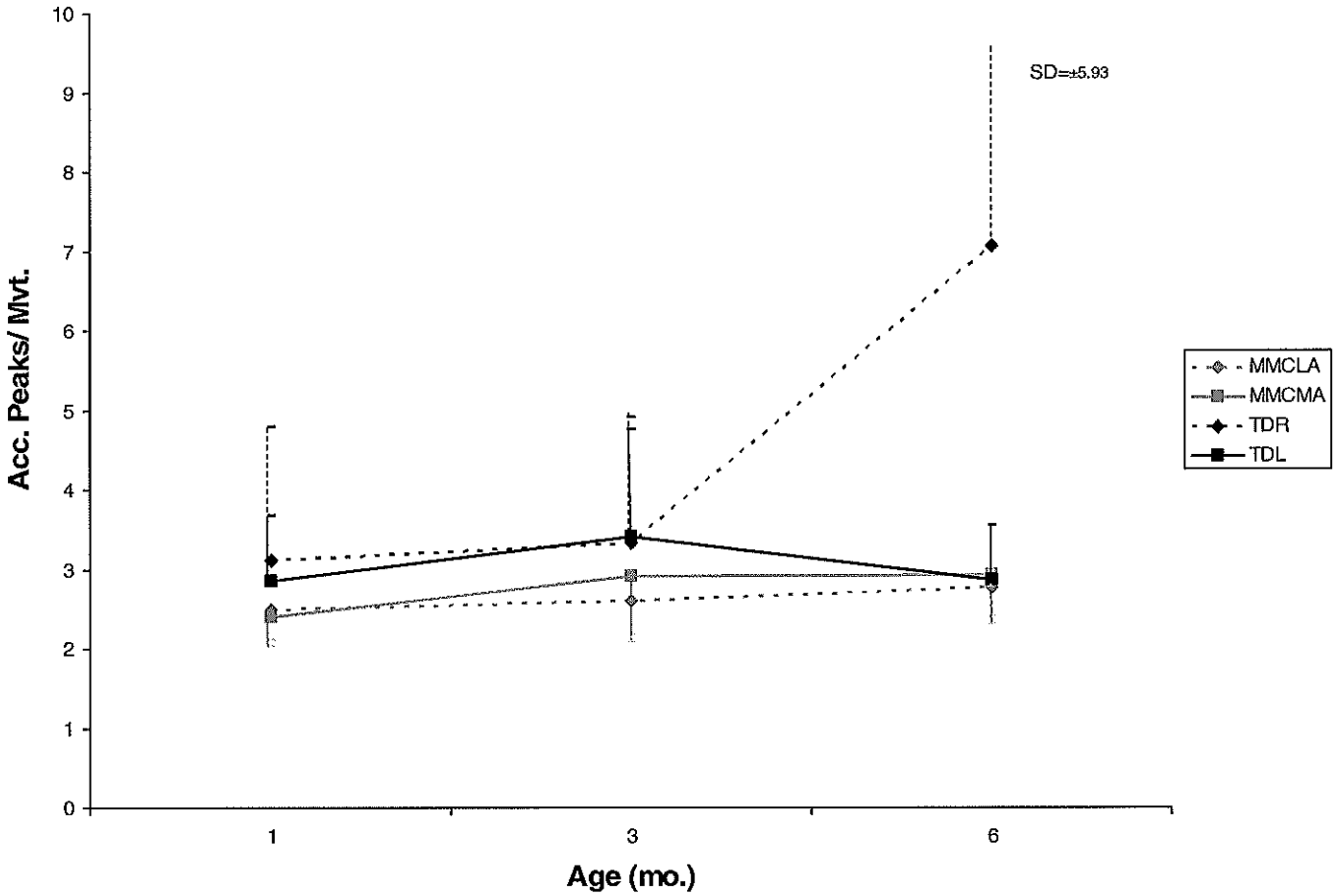

Fig. 6. Acceleration peaks per movements of the right and left legs of infants with typical development (TD) and the more (MA) and less affected (LA) legs of infants with myelomeningocele (MMC).

significant effects. The mean values shown in Figure 6 suggest that infants with TD consistently produced more acceleration peaks per movement than infants with MMC; infants with TD reached a peak mean of 3.35 at 3 months of age whereas infants with MMC maintained a fairly stable mean of approximately 2.5 across this age range.

DISCUSSION

Overall, our results suggest that young infants with MMC show depressed leg activity compared with their peers with TD. However, the profile of their behavior is complex and shows fewer differences than might be expected from other published reports as well as differences not previously reported.

For example, Chapman[20] and Sival et al[19,22] reported that infants with MMC produced significantly fewer leg movements than their peers with TD. However, our main effect for group did not reach statistical significance. The group means looked quite different (Fig. 2); however, the variability in our sample was high, and thus, we may have lacked power to obtain statistical significance. Only the more differentiated group by leg interaction reached statistical significance, suggesting that the frequency with which infants in these groups moved differed across legs. In this case, infants with TD moved one leg considerably more than the other whereas infants with MMC move both legs at a similar rate.

Beyond power, the fatigue factor may have played a role in our lack of a significant group difference. Before testing infants in the spontaneous movement context we engaged them in upright activity of 2 other types. We attempted to elicit (a) newborn steps and (b) stepping patterns on a motorized treadmill. One could imagine that engaging infants in activity primed them to continue to be awake and active; we also allowed rest periods between trials and contexts. Although, one might also argue that the metabolic cost of this earlier activity may have reduced their energy reserve and subsequent activity level. When we compared our infants' rate of movement with that of Chapman's[20] youngest infants (18 weeks), our rates seem to be depressed somewhat. He reported 1.38 movements/sec for infants with TD and 1.2 movements/sec for infants with MMC compared with our rates of 0.83 and 0.72, respectively. Further, Chapman's group effect was based on overall behavior across 3 contexts: supine and seated in two different infant seats. Mean differences in leg movement frequencies were smallest during the supine context. Thus, group differences in supine spontaneous leg movement may be smaller than in some other contexts, but real; our sample may have failed to achieve statistical significance because of high variability across infants combined with overall activity levels depressed by fatigue.

One final factor that must be considered is developmental trends. Over the first year of postnatal life infants

with TD have been shown to increase and decrease frequencies, patterns, and quality of leg movements.[12,13,16] For infants with MMC, Chapman[20] found an increase in frequency of leg movement from ages 18 weeks through 26 weeks followed by a reduction in frequency by 30 weeks. Sival et al[22] reported a reduction in leg movements over the first few postnatal days of life in infants with MMC. Our sample was older than Sival et al's very young infants and overlapped a bit with Chapman's. It is possible that the developmental trajectory for infants with MMC is nonlinear, with a reduction in leg activity after birth because of surgical intervention, followed by high variability and more overlap with infants with TD for several weeks, with clear group divergence emerging over developmental time.

Although leg movement frequency showed a complicated pattern of results, the results for duration of movement were more straightforward. Infants with TD produced movements of longer durations than infants with MMC and both groups increased movement duration with age. Together these results suggest greater strength or energy reserves in the babies with TD, that is, when infants with TD initiated a movement they maintained that movement longer. Conversely, this argument proposes that infants with MMC were weaker than their peers yet growing stronger with age, perhaps at a reduced pace.

We specifically addressed activity for each leg because we anticipated that the impact of the neural tube defect would not, in many cases, be uniform across limbs.[21] At the joint level alone infants with MMC often show asymmetries in their need for surgeries, presence of club foot, hip dysplasia, and so on. Yet, in most studies of infants with MMC the movement of both legs is simply pooled. Somewhat surprisingly we observed greater asymmetry in our infants with TD than those with MMC. Observing asymmetry in infants with TD was not, by itself, surprising. Over the first few months after birth infants with TD show a fairly common developmental trajectory in which early coupled (alternating right and left) leg activity gives way to more independent leg and joint actions.[13,23,24] In doing so, infants become able to convert spontaneous kicking movements into instrumental behaviors.[25,26]

Several recent studies suggest that persistent symmetry in leg movements and difficulty adapting their leg patterns may reflect underlying neural control challenges. Heathcock et al[27,28] used a reinforcement paradigm to encourage 2- to 4-month-old infants born prematurely and infants born with TD to shift from alternate kicking to single-leg kicking. When one leg was reinforced for activity infants born prematurely increased the activity of both legs, rather than just that of the reinforced leg as was the response in infants with TD. These results led the authors to propose that infants born prematurely show signs of difficulty learning to adapt their spontaneous movements, and to differentiate their limb movements. In a related study, Sival et al[22] reported that young infants with MMC, compared with peers with TD, showed fewer isolated leg movements. They coded general movements (GM) and isolated leg movements. Their data suggested that infants with MMC were more likely than their peers to move their lower limbs concurrently with other limb movements than infants with TD.

Traces of concern regarding delayed emergence of leg control may also be evident in our data reflecting more frequent acceleration peaks in infants with TD than MMC. In the motor learning literature, increased accelerations during a movement tend to suggest corrections, and thus, poorer control. However, very early in development the opposite may be true, in that it may reflect the capacity to explore and to begin to modulate movements, rather than simply letting them 'run their course' once initiated. Hopkins et al[29] describe a notable developmental change in the spontaneous movements of infants with TD around 2 to 3 months of age, from slow 'writhing' movements to ones with a more 'fidgety' quality. Hadders-Algra et al[30] suggested this transitional stage may be associated with changing from greater coactivation to reciprocal activation of muscles.

In our study, infants with TD showed asymmetry in leg activity but also a greater tendency for the right leg to demonstrate more movements than the left leg. The significance of this specific right preference is unclear. Across studies of the emergence of limb preferences (primarily arm/hand), when preferences occur they seem to favor the right limb, but fluctuations in preference are common and limb differentiation seems not well established before 6 months, particularly for the legs.[31–35] At this point we propose the relevance of our results lies in the group difference, that is, in reinforcing the uniquely different, atypical, developmental trajectory emerging for infants with MMC compared with their peers. As the sequential behaviors marked by early symmetry, followed by asymmetry, and ultimately limb preference emerge over time they reflect developmental progress and enable the child to explore and develop ever more complex and refined control. Depressed activity levels may reflect inherent disruption to the neuromotor pathways and, like a vicious cycle reduces the opportunities for infants to learn to control their movements and to build the neural and muscle organization and cooperation needed to acquire functional skills.

CONCLUSIONS

Infants with MMC, across ages 1 to 6 months postbirth, show depressed spontaneous leg activity, compared with their peers with TD. However, their behavior is not simply and uniformly less; they also demonstrate qualitative differences that suggest a unique developmental trajectory of the development of control.

Movement durations were shorter for those with MMC but both groups maintained movement longer as they grew older. Leg movement frequency showed that infants with MMC moved both legs at similar rates whereas their peers with TD moved one leg more than the other, and particularly more than infants with MMC. By differentiating between legs, rather than pooling leg data across variables, we were also able to observe a recurring presence of differences between the groups in their symmetry and

asymmetry of leg control. Infants with TD showed greater evidence of the capacity to differentiate their limb movements suggesting increased capacity to explore and discover how to use their legs functionally. We propose the need for further research to determine if increasing the frequency of early leg movements, via therapeutic intervention, will enable infants to increase durations and frequency of spontaneous movement and facilitate development of functional control.

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