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Impact of Enhanced Sensory Input on Treadmill Step Frequency: Infants Born With Myelomeningocele

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Abstract

Purpose—To determine the effect of enhanced sensory input on the step frequency of infants with myelomeningocele (MMC) when supported on a motorized treadmill.

Methods—Twenty seven infants aged 2 to 10 months with MMC lesions at or caudal to L1 participated. We supported infants upright on the treadmill for 2 sets of 6 trials, each 30s long. Enhanced sensory inputs within each set were presented in random order and included: baseline, visual flow, unloading, weights, Velcro and friction.

Results—Overall friction and visual flow significantly increased step rate, particularly for the older group. Friction and Velcro increased stance phase duration. Enhanced sensory input had minimal effect on leg activity when infants were not stepping.

Conclusions—Increased friction via Dycem and enhancing visual flow via a checkerboard pattern on the treadmill belt appear more effective than the traditional smooth black belt surface for eliciting stepping patterns in infants with MMC.

Keywords

age factors; ambulation; body weight; early ambulation; infants; child development/physiology; gait disorders/neurologic; lumbar vertebrae; meningomyelocele; motion perception; motor activity; proprioception; treadmill test; weight bearing

INTRODUCTION

Myelomeningocele (MMC) is the most common neural tube defect in the US.¹ Although the incidence of neural tube defects has decreased considerably in the last decade with the introduction of folic acid and improved prenatal care, approximately 1,500 to 2,000 of 4 million live births still present with spina bifida.² The physical health related quality of life for individuals with MMC remains significantly lower compared to individuals with typical development (TD), particularly for children who are not ambulatory.³

Whereas spinal cord lesions emerge very early in gestation, physical stimuli also surround the embryo and fetus and during this period, and babies with MMC move as frequently as those with TD.⁴ This may be because they are stimulated by movement of the surrounding fluid systems, sporadic uterine contractions or maternal motor activity.⁵ The neonate enters a world with less continuous extrinsic motor stimulation which may partially account for the diminished movements observed in infants with MMC during the first weeks of life.⁶ Recovery from spinal surgery in addition to possible shunt surgery will also contribute to reduced movement in the newborn with MMC. Additionally, greater muscle force is required for all neonates to generate anti-gravity movements since they no longer have the advantage of buoyancy provided by amniotic fluids but this is particularly taxing for babies with MMC. Development of control of the lower limbs is also difficult, given that most lesions occur in the lumbar and sacral regions from which nerves arise that innervate the pelvis and legs. Infants with MMC who develop locomotor skills begin to walk on average 2 years later than infants with TD.⁷ The presence of club feet, dislocated or subluxed hips and muscle contracture will further impede the onset of ambulation. Approximately 20% of infants with a high lumbar level lesion, 80% of infants with a low lumbar level lesion and over 90% of infants with a sacral level lesion achieve independent walking.^{7, 8} In addition to lesion level, spasticity, poor balance and the number of shunt revisions add to a poor prognosis for independent walking.^{9, 10} The kinematics of gait in children with MMC differs from that of children with TD. For example, children with MMC show more trunk rotation, anterior pelvic tilt and increased pelvic movements.¹¹ The majority of ambulatory children with MMC walk with aids; many also lose the ability to walk or choose not to walk independently during late childhood.⁸ Health risks are also associated with reduced ambulation and gait problems, such as lower physical activity and increased risk for obesity.^{3, 11, 12, 15, 16}

Improving the activity and gait outcomes for children born with MMC depends on creating a strong foundation of early motor strength and control of pelvis and legs. Currently there is no general agreement on best management of infants with MMC, unlike traumatic spinal cord injuries. For such cases, accepted practice is that gait therapy is initiated as quickly as possible after surgical repair to facilitate the recovery of damaged and development of new neural pathways.¹⁷ Early introduction of rehabilitation provides a better prognosis for independent ambulation for adults with spinal cord injury.¹⁷ While the mechanisms of spinal cord injuries due to trauma versus neural tube defects are different, as are the contexts in which they occur (well-developed neuromuscular system or very early in development) one important process for improving neuromuscular control is the same, the need to move the limbs in as close to functional patterns as possible.¹⁸ But how does one engage infants with developmental spinal cord injuries in leg activity that promotes strength and neuromotor control and that is functionally relevant to walking?

One potential option is presented by research conducted with infants with TD and those with Down syndrome in which babies were supported upright on a pediatric treadmill. Practice stepping on the treadmill, 3 to 5 times a week in their homes, resulted in significantly increased step frequency and led to earlier onset of walking for those with Down syndrome.^{19, 20} We know that new skills and neuromotor control develop through exploration and the repetitive process of moving, or acting, and perceiving the consequences of that effort, particularly in a functional context.²¹ Increasing the frequency of motor output and afferent input to the nervous system stimulates the development of new networks through the growth of dendritic connections, changes in sensitivity of neural membranes and alterations in neurotransmitters.^{22–24} In infants, neural plasticity is maximal, and therefore early therapeutic intervention is imperative to employ this feature of neuroplasticity.^{25, 26} Studies indicate that lower motor neurons caudal to the lesion are indeed functional and hence have the potential to respond to increased afferent input, as Geerdink et al.

demonstrated when lumbar magnetic stimulation was applied to segments caudal to the lesion.²⁷ However, the same study reported that transcranial stimulation did not produce any discernible motor activity.²⁷ Stark and Baker (1967) theorized that there were different levels of neurological functionality of spinal cord segments caudal to the lesion, ranging from a normal cord, a partially functioning cord, an isolated functioning cord to a functionless cord.²⁸ An added impetus to commence treatment early is that muscle cells have the greatest potential to hypertrophy at a young age when protein accretion rates are highest.²⁹

Infants with MMC exhibit less spontaneous movement than infants with TD, resulting in less opportunity than their peers with TD have to forge organized motor patterns, such as those needed for walking.³⁰ However, previously we demonstrated that infants with MMC will respond to being supported on a motorized treadmill by producing steps although at a lower rate than those performed by infants with TD.³¹ This activity could, ultimately, be tested for its capacity to assist babies with MMC in building the strength and control needed to walk. The use of a treadmill to promote the stepping response in infants may present itself as an early therapeutic intervention exploiting the high degree of neuroplasticity previously described. But, to do so we must be confident that the context optimizes the potential for babies to improve. Ulrich & colleagues³² showed that modifications made to the baseline treadmill context could significantly increase step frequency in infants with Down syndrome, if presented during periods when stepping on the treadmill was low or unstable. Here we followed that strategy, testing infants with MMC at chronological ages during which their treadmill stepping responses were relatively low. Further, our modifications targeted sensory systems particularly relevant to this specific population, tapping into their existing capacities, enhancing the input to sensory systems that are compromised, and attempting to maintain the sole of the foot in contact with the treadmill belt in order to increase the sensation of motion.

Our purpose in this study was to determine if enhancing the sensory input to infants with MMC would increase their step frequency over baseline. The specific enhancements we used were: (1) visual flow, (2) abrupt unloading of ankle and hip joints in stance, (3) increased mass – increased input to pressure receptors in the foot, (4) Velcro sock – increased time in stance, and (5) friction (Dycem®) – during stance

METHOD

Participants

We enrolled 11 infants aged 2 to 5 months and 16 infants aged 7 to 10 months corrected age, with MMC at, or caudal to the level of L1. We chose these 2 age groups to represent distinct periods during the first year related to development of motor patterns. The 2 to 5 month period is when milestones for upper body control are more commonly achieved and the 7 to 10 month period tends to include increasing numbers of trunk, pelvis, and lower body control behaviors.³³ We excluded infants who had neuromotor abnormalities other than those associated with MMC (e.g. Arnold Chiari II, hydrocephalus) or if they had a gestational age at birth less than 28 weeks. We recruited infants through fliers and spina bifida clinics in hospitals in southeast Michigan, northeast Ohio, and southeast Wisconsin. Approval for the study was granted through the Institutional Review Board at the University of Michigan. Parents provided written informed consent for their infants to participate in this study and completed a medical status and history survey (including shunt status, level of spinal fusion surgery and subsequent surgeries). Table 1 presents participant characteristics.

Procedure

We conducted the testing in the Developmental Neuromotor Control Laboratory (44%) or to increase sample diversity and numbers, we accommodated families by offering to perform all testing in families' homes (56%). To prepare infants for testing we removed clothing and placed reflective markers (8mm diameter) on the iliac crest, greater trochanter, lateral knee joint line, lateral malleolus and ventral surface of the third metatarsophalangeal joint of the right lower extremity. We positioned bipolar surface electrodes over the muscle bellies of the lateral gastrocnemius/soleus (GS), tibialis anterior (TA), quadriceps femoris (QUAD) and lateral hamstring (HAM) muscles on both legs. We placed a ground electrode over the right patella. We recorded EMG signals via 8 channels of a Myosystem 1400A (Noraxon Inc, Scottsdale, Arizona) unit at 1000Hz.

The custom-designed treadmill (Carlin's Creations, Sturgis, MI, USA) was 18cm high, 42cm wide and 82cm long and we placed it on top of a table 73cm high. We used two digital camcorders (Panasonic PV-GS 35) recording at 60Hz to document the position of the infants' lower limbs. We positioned camcorders on tripods lateral to the right side of the baby, the optical axes forming an angle of 70° to 90°. We calibrated the camcorders prior to testing with a small calibration frame (Peak Motus) and synchronized them with a signal. We also used the same signal to synchronize the EMG with the camcorders.

We manually held infants upright so that their feet rested on the belt of the treadmill in a partial body-weight-supported position for twelve 30-second trials. The treadmill testing lasted a total of 6 minutes, with rest intervals in between trials. Some of the weight of the infant was supported by the tester, thereby subjecting the infant's lower limbs to varying amounts of pressure due to their individual differences in weight acceptance and degree of inward or outward foot rotation (e.g., club foot). The tester observed the foot position of the infant and increased support if there was prolonged lateral contact of the foot with the treadmill surface, which was observed to occur in 48% of infants with club foot and 11% of those with structurally normal feet. Trials were presented in 2 sets of the 6 conditions listed below; within each set conditions were presented in random order. We set the belt speed at 0.16 m/s because it proved to be an optimal speed in our previous longitudinal study of treadmill stepping in infants with MMC.³¹ Conditions were: (1) baseline – smooth black belt surface; (2) visual flow – black and white checker-board patterned belt used to elicit visual flow sensation; this condition was selected because research shows infants often respond to visual flow that suggests movement by initiating trunk or limb movements³⁴; (3) unloading – infant was held near the end of the treadmill, so the child's feet abruptly dropped off the surface, rapidly unloading the hip and ankle joints³⁵; biomechanists propose that unloading at the hip and ankle activates joint receptors that cause motor units to fire and initiate the swing phase of stepping³⁶; (4) weights – attached to the infant's shanks, tailored to their individual weight and equal to approximately 50% of shank mass; we wanted to enhance the normal contribution of passive pendular motion in swing phase and to increase the input to pressure receptors in the foot during stance phase³⁷; (5) Velcro – infants wore socks with strips of Velcro sewn on and the treadmill was covered with a felt-like material; Velcro maintains the infant's foot in contact with the treadmill surface for a longer period during which time the extensor muscles are expected to be active; greater contraction of the flexor muscles is necessary to draw the foot away from the treadmill; Ulrich and colleagues³² demonstrated that Velcro resulted in the greatest number of steps for infants with Down syndrome; (6) friction – belt made of Dycem® (a non-slip surface); the goal was to limit the infants' feet from sliding on the surface and not moving backwards with the belt, as we observed in previous studies when infants did not support much of their own weight.

Subsequent to treadmill training we took anthropometric measurements including body length, weight, greater trochanter to lateral malleolus length, thigh length, foot length, thigh

circumference and leg circumference. The purpose of these measurements was to determine whether some were a factor in the stepping response and also for normalization and further analysis in future studies. We recorded aspects of the infant's medical history including lesion level, surgeries and musculoskeletal conditions. We assessed concurrent motor skill development level by administering the motor items from the Bayley Scales of Infant Development III.³⁸ Table 1 presents medical and anthropometric characteristics of the participants.

Data Reduction

We behavior coded the videotapes to determine stepping patterns. Frame-by-frame analysis of each trial was made with Peak Motus Version 8 software. The coders (n=4) had to achieve a coefficient of agreement of 0.85 (interobserver reliability coefficient, kappa) through comparison of their work with that of previously validated coders for the same set of trials by using training tapes. We extracted the following parameters from the videos.

Interlimb stepping patterns—Four types of steps: alternating (a step preceded or followed by a step of the contralateral limb with overlap), single (not preceded or followed by a step of the contralateral limb), parallel (both legs swing forwards simultaneously) and double ('stutter' step within a sequence of alternating steps).

Step rate—Total number of steps per trial divided by the trial duration in seconds.

Step events—Video frame number for toe-off, touch-down and end of stance for alternating steps and single steps. We normalized the total cycle, stance and swing durations calculated from these events by dividing the parameters by $(l_o/g_o)^{1/2}$ where l_o is the segment length and g_o is standard gravity, 9.81ms^{-2} .³⁹

Leg activity rate (LAR)—We coded leg activity during frames in which no steps were present, every 5 seconds with dichotomous values: 0 = no leg movement and 1 = clear leg movement. We divided the total points accumulated by the number of frames and multiplied the index by 60 to obtain the LAR.

Ponderal index (PI)—Indicator of body proportion commonly calculated for infants.⁴⁰ We calculated the PI using the formula: $PI = (\text{Weight(g)} / \text{Length(cm)}^3) \times 100$

Data Analyses

We used SAS version 9.2 (SAS[®] Inc., Cary, NC) for statistical analyses. We applied the mixed model procedure to conduct analyses of variance (ANOVA) with repeated measures. We performed pairwise comparison tests to determine significant differences between parameters. We set statistical significance a priori at $p < 0.10$ due to the high performance variability in this population, the wide range in ages, lesion levels and shunt surgery, cerebellar malformations and orthopedic complications. We have presented statistics mainly for those results that achieved statistical significance and on occasion for those without statistical significance where we consider this lack of significance to be of interest. The lesion groupings were low-level lesions (L5 and caudally) middle-lesion level (L4) and high-level lesions (L1–L3). The rationale for these divisions is based on the likelihood of the infants becoming community walkers into adulthood.^{31, 41}

RESULTS

We organized our results into two sections. In Part 1 we provide a simple overview of the overall motor development status of this sample and the role of PI and age in days in step

and leg movement rates. We list the values of the Bayley Raw Motor Score and PI in Table 1. In Part 2 we focus on the main questions of interest, the enhanced sensory input on step rate and quality and leg activity.

Part 1: Developmental Overview

Bayley Motor Scale—We used the raw total scores (number of items passed) to reflect each infant's motor performance on the Bayley Scale. Overall, the motor scores increased significantly with age as shown in Figure 1, with a steeper increase for infants with a low-level lesion compared to those with a high-level lesion. While the mid-level lesion sample was too small and narrowly defined by age to conduct a regression analysis, Figure 1 suggests less clear change in motor outcomes related to age for this subgroup. The increase in Bayley score with age was less pronounced than that we previously observed for infants with TD who also displayed a higher R-squared value. We divided the Bayley score into 5 levels (10 to 19, 20 to 29, 30 to 39, 40 to 49, and 50 to 59) and applied a 5 (Bayley score) $\times$ 6 (condition) ANOVA to examine the effect of Bayley score and condition on total steps produced with repeated measures on condition. We found no significant relation between concurrent motor skill performance and step response reflected in the Bayley scores.

PI and Step Rate, LAR—The relation between PI and overall step rate is illustrated in Figure 2. We observed a trend for step frequency to decrease as PI increased, but this was true primarily for infants with middle- and low-level lesions, with no obvious effect for those with high-level lesions. We observed a decrease in LAR as PI increased for infants with a low-level lesion (Figure 3). However, for infants with middle- and high-level lesions the opposite trend emerged, LAR increased as PI increased.

Step Types Produced—Both the younger and older age groups generated all 4 types of stepping patterns (Figures 4A and 4B). However, for younger infants the predominant step type was single, followed by parallel, alternating, and double. With age, alternating more than doubled in percent of all steps, as did parallel steps, while single steps reduced by half and double steps nearly disappeared.

Part Two: Context Effect on Step Frequency, Temporal Parameters, and LAR

Because there were, particularly for the younger infants, only a small number of steps in each of the 4 step categories for each of the conditions, we pooled the step types together to increase the power of subsequent statistical tests.

Step Frequency

Absolute Step Rate: To examine the effect of age, lesion level and conditions on total steps produced we used a 2 (age) $\times$ 3 (lesion) $\times$ 6 (condition) ANOVA for repeated measures on condition. We found a significant age effect ($F_{1,23} = 6.76, p = .016$), a significant lesion level effect ($F_{2,23} = 3.13, p = .063$), a significant condition effect ($F_{5,115} = 2.38, p = .043$), a significant age $\times$ condition interaction ($F_{5,115} = 3.13, p = .011$) and a lesion $\times$ condition interaction ($F_{10,115} = 1.99, p = .040$). Figure 5 plots step rate by age and condition, within each lesion level, A (low), B (middle), and C (high). These figures illustrate that the significant age $\times$ condition effect reflects that while conditions effectively increased step responses over baseline, the level of effect varied by age, showing a greater effect for older infants than younger ones. Further, the effect of conditions presented was different across lesion levels, being greater for low than high lesion levels. For those in the middle-lesion group, no conditions showed clear improvement over baseline, though the age effect could not be examined because our sample included only infants aged 7 to 10 months.

To examine more closely the age by conditions interaction we compared first the preferred conditions of younger babies to those of older babies. For younger infants, friction produced the greatest increase in step rate (0.16 steps/s $\pm$ 0.18) compared to the baseline condition (0.11 steps/s $\pm$ 0.10), although the paired comparison post hoc did not reach statistical significance. For older babies, visual flow elicited the most steps (0.26 steps/s $\pm$ 0.23) compared to the baseline condition (0.19 steps/s $\pm$ 0.15), but both visual flow and friction achieved statistical significance: visual flow (DF=75, $t=-2.37$, $p=.021$) and friction (DF=75, $t=-1.92$, $p=.058$) compared to baseline for the older group.

To examine the lesion level by condition interaction we began by conducting a post hoc 2 (age) $\times$ 6 (condition) ANOVA with repeated measures on condition for the low-level lesion group. We began here because, as Figure 5a illustrates, this subgroup had both the most participants (and thus statistical power) and the largest overall step response. Results showed a significant condition effect ($F_{5,55}=2.21$, $p=.066$), age effect ($F_{1,11}=5.87$, $p=0.034$) and an age $\times$ condition interaction ($F_{5,55}=2.68$, $p=.031$). Figure 5a suggests that whereas for older infants all enhanced sensory conditions produced more steps than baseline, for younger infants only friction seemed to elicit more steps. Post hoc paired comparison tests among conditions showed statistically significant results for visual flow (DF=60, $t=-1.71$, $p=.092$) and friction (DF=60, $t=-2.81$, $p=.007$) compared to baseline.

Figure 5b suggests that for infants in the middle lesion level (L4) no enhanced sensory conditions improved their step frequency, though we observed a statistically significant decrease in responses for unloading (DF=25, $t=2.35$, $p=.027$) and weights (DF=25, $t=2.16$, $p=.041$) compared to baseline.

Infants with high-level lesions responded more to friction (0.10 steps/s $\pm$ 0.05) compared to baseline (0.09 steps/s $\pm$ 0.05) though visual flow showed also a slightly higher mean response compared to baseline for the older infants only. However, these changes did not achieve statistical significance.

Normalized Step Rate: The variability in mean step rate across infants was high, ranging from 0.03 to 0.67 steps/s. Therefore, we normalized step rates across conditions for each infant to his/her own mean. Normalized responses (see Fig. 6) show more clearly that, generally, visual flow and friction were the conditions most effective in eliciting steps when infants with MMC are supported on a pediatric treadmill. We plotted normalized responses as a function of age and lesion level. For the youngest infants only friction increased step rate over baseline. For those with middle level lesions no conditions significantly increased step response compared to baseline; unloading and weights, in particular significantly reduced step responses.

Step Cycle Effects—To determine the effect of conditions on step cycle duration we used a 2 (age) $\times$ 3 (lesion) $\times$ 6 (condition) ANOVA with repeated measures on condition. Cycle duration, normalized to leg length, was the dependent variable. We found significant age ($F_{1,6}=6.77$, $p=.041$) and condition ($F_{5,15}=4.83$, $p=.008$) effects. Figure 7 shows that, with age, normalized cycle durations reduced across all conditions. The absolute cycle durations were higher in the infants with longer legs and they demonstrated increased strength of control; when normalized to leg length the cycle duration decreased. The effect of condition was less clear cut, though overall, Velcro tended to increase cycle duration while unloading tended to decrease cycle duration. An 2 (age) $\times$ 3 (lesion) $\times$ 6 (condition) ANOVA with repeated measures on condition for swing and stance phase durations resulted in statistically significant effects for stance phase durations for condition ($F_{5,15}=3.65$, $p=.023$).

LAR—Figure 8 illustrates the very high variability across infants in the LAR to all conditions and across age groups and lesion levels. When tested in a 2 (age) $\times$ 3 (lesion) $\times$ 6 (condition) ANOVA for repeated measures on condition with LAR as the dependent variable, we obtained only a significant age effect ($F_{1,23} = 4.35$, $p = .048$). Older infants produced more leg activity when not stepping than younger infants. Nevertheless, the comparison to Figure 5 shows some parallels that may guide selection of conditions that elicit maximal action. For example, Figure 8a suggests that for older infants with low-level lesions visual flow and friction seem to work better than baseline. For young infants this is limited to friction. All conditions reduced the response compared to baseline for infants with middle-level lesions, with unloading and weights showing the most marked effects. For infants with high level lesions, in both age groups, little improvement was evident across enhanced sensory conditions, though notably none of these conditions appears to reduce leg activity compared to baseline. We found for the infant group as a whole that the condition of unloading decreased LAR from $5.88/s \pm 6.40$ at baseline to $3.65/s \pm 3.79$. However, no condition produced a marked increase in LAR.

DISCUSSION

Our purpose for this experiment was to determine if we could enable infants with MMC to increase their step rate when supported on a pediatric treadmill by enhancing the sensory input during test trials over that of typical treadmill belt conditions. Overall, our results show that we can, and that of the 5 sensory enhancements we created, friction and visual flow worked optimally to increase step frequency.

We chose to increase friction by covering the treadmill belt with Dycem to avoid a response observed in some infants with MMC in our previous work.³¹ That is, if they accepted minimal weight on their feet or foot contact was on the side or ball of the foot, their feet moved backward with the belt only a small amount and steps were less likely to result. We used Dycem to reduce slippage. In this context, it increased the opportunity for the treadmill dynamics to be effective. When the foot touched down it created a more stable base for greater weight acceptance on the foot and kept the foot in contact longer. When leg joints and muscles are extended and then unloaded at the end of stance passive pendular dynamics and viscoelastic muscle properties can assist with forward motion and joint flexion.

Visual flow has been shown by numerous researchers to elicit motor responses in infants.^{34, 42, 43} We chose this condition to access a sensory system that was not diminished by the neural tube defect and thus could, and did, facilitate infants' initiation of body movement. However, this condition was less effective for younger babies than older babies. This may have been because younger babies failed either to “perceive” the motion implied or simply failed to attend to it. Although we did not specifically assess babies looking time, Moerchen and Saeed⁴⁴ showed that babies with typical development who spend more time looking directly toward the belt in this context, rather than looking away or closing their eyes, stepped more frequently.

Like friction, Velcro also helped maintain greater foot contact after touchdown and this was the condition Ulrich et al.³² found to be optimal for infants with Down syndrome. The difference here seemed to be the strength required to pull their feet off the belt, which was particularly low for young babies with MMC and those with high lesion levels. Similarly, adding weights may have increased proprioceptive input but these babies simply didn't have sufficient strength to overcome this added mass.

The responses we observed to friction and visual flow were affected as well by age and lesion levels. Infants who were in the older group stepped more frequently and responses

increased as group lesion level lowered. In fact, those infants in the older age group with the lowest lesion levels responded more to all enhanced sensory conditions than to baseline. The question remains, why was the effect on younger infants and those with higher lesion levels so small? We propose 2 factors. First, these infants responded to baseline with a low number of steps, a finding we had demonstrated in a previous study.³¹ These are the most fragile infants of this population, for whom lifting the legs and stepping may be a particularly difficult and energy demanding response. Thus, expecting any real-time modification that does not reduce the muscle force demands may be unrealistic. More effective may be efforts to increase strength first before expecting infants to use their muscles and step. Second, our design allowed infants only 30 seconds to adjust to each new test trial and we moved fairly quickly from one trial to the next. We did this to avoid tiring infants and allow examination of several enhanced sensory conditions, since all data were collected on a single day. Unfortunately, by doing so, we may not have allowed this inherently unstable response to settle into each new sensory enhancement before the trial ended. Future studies should consider examining the most promising conditions by presenting them for longer periods of time, and comparing responses of stronger infants, those who generate more extensor force when supported on the treadmill, with those less able to do so. Additionally, over 37% of our infants presented with bilateral club feet. These infants made initial contact with the lateral aspect of the foot in 47.2% of cases which reduced during midstance. However, even those infants without clubfeet made initial lateral foot contact in 19.6% of their steps, compared with about 8% lateral foot contact for infants with TD with structurally normal feet. The effect of clubfoot on the posture was to reduce the treadmill surface area in contact with the foot in addition to limiting weight bearing through the lower limbs, with the tester subsequently providing more bodyweight support. This decrease in sensory enhancement in infants with clubfeet may account for the reduction in number of steps elicited overall in infants with MMC.

Interestingly, in this study we observed an age effect that was only minimally observed toward the end of the first year in our previous longitudinal study.³¹ We believe this occurred because in the previous study only the baseline condition was used. Here, by increasing the opportunities for sensory input to engage the motor system, greater overall stepping emerged with age.

As a group, infants with middle lesion levels differed in several respects from the pattern shown by the lower and higher lesion level groups. They did not increase step frequency in response to any enhanced sensory input conditions, and step frequency diminished during unloading and weight conditions. We do not have a definitive argument for this divergence, but one contributing factor may be the very small subsample size, coupled with its very high variability. The middle lesion level group was represented by half as many participants as the other two groups, and was comprised of only 6 infants. Individual profiles showed that 3 of the infants in this group showed means for visual flow and friction that were equal to or higher than their baseline means. For only 2 babies in this group did the baseline the condition produce the most steps and for only 1 of them was that number more than 25% higher. Overall, in such a small sample, these less common response patterns can overwhelm a group mean. A larger sample of infants with an L4 lesion level diagnosis will be needed to examine this subsample behavior more definitively.

We did not observe a significant increase in leg activity beyond steps, as a function of enhanced sensory input, although the means for friction and visual flow were a bit higher than other conditions, paralleling the more significant outcomes we found for step rate under these conditions. Previously we showed that when infants with MMC were supported upright on a moving belt that was like our baseline belt in this study, infants moved their legs more even when not stepping than they did when supported upright on the treadmill and

the belt did not move. The explanation for this lack of increase is not obvious, although perhaps when increased sensory flow reached a sufficient level to stimulate activity, this presented as a patterned motor response, a step, rather than random leg activity.

As we observed previously the step patterns infants with MMC produced at younger ages tended to be dominated by single steps and these decreased with age, while the proportion of alternating steps increased with age.³¹ The amount of increase in alternating steps was less than expected from previous work, to about 20% here and 40% previously. We propose that the increase in response frequency may first come in the form of the simplest patterns, single, or parallel steps, and require greater control and practice to more consistently respond with alternating steps.

Our results also showed a negative correlation between PI and step rate for the low- and middle-level lesions. A higher PI indicates increased adipose tissue for body size. Luo et al.⁴⁵ also reported a negative correlation between number of alternating steps and percentage of total body fat as determined by skinfold thickness. Our interpretation for our findings is that infants with increased adipose tissue per body size have more difficulty initiating step cycles, due to the relatively heavier lower limb without concomitantly greater muscle force. For infants with high lesion levels who stepped significantly less, we observed no effects of PI.

We found no statistical relation between infants' step frequency and their scores on the Bayley Scale of Infant Development (BSID). This may be because the BSID is heavily weighted toward upper limb and trunk motor skills, and thus, may be insufficiently sensitive to neuromotor delays primarily occurring in the lower limbs. Perhaps a scale such as the Alberta Infant Motor Scale⁴⁶ would have identified a relation. Alternatively, the capacity for the dynamics of the treadmill to assist and elicit stepping patterns may occur before babies are able independently to create the functional motor patterns of sitting, crawling, and cruising. Previous studies provide limited insights. For example, Thelen and Ulrich⁴⁷ found no relation between BSID scores and the onset of improved treadmill stepping in infants with TD, whereas Ulrich et al.¹⁹ found for babies with Down syndrome that specific BSID items were associated with improved treadmill stepping, but not an overall score. In future studies we will investigate this issue further by employing a more sensitive set of items to reflect concurrent levels of lower limb function (skill).

Although our results show that enhancing sensory input can increase motor output and that infants with lower level lesions and older infants responded to this input more than young infants and ones with higher lesions, the variability among infants must also be emphasized. Scatterplots and standard deviations illustrate this, as does the very high variability among infants in step rate. These reflect that behavioral outcomes are influenced by many factors, including other complications due to this neural tube defect, such as club feet (reduced contact area), hydrocephalus, or hip subluxation/dislocation, degree of damage to ascending and descending neural tracts, level of medical care and physical therapy, as well as family support networks.

It is important to note that in the future, the possible use of treadmill practice for early intervention for children with MMC would need to be paired with close monitoring of its effect on muscle imbalance around the hips and knees. Infants and children with MMC, especially those with lumbar lesions, are prone to show muscle imbalances in this region. Thus, careful attention to pelvis, hip, and spine alignment would be critical in order to avoid the potential development of muscle contracture and bony deformities.

LIMITATIONS

In this study we presented each enhanced sensory condition for only 30 seconds and moved infants quickly from one context to another. Future studies should examine the effect of longer exposure time to allow infants to adapt to each context. Further, we enrolled only a small number of infants when sorted by age group and lesion levels; the mid-lesion level subgroup included only older infants and half as many in total as the lower and higher lesion level subgroups. Future studies of this nature should increase the sample within subgroups. We utilized the same sensory enhancements for the youngest and oldest infants. Future studies should test if other modifications to the treadmill context could improve responses for younger infants and those with the highest lesion levels.

CONCLUSIONS

Our results showed that when supported upright on a pediatric treadmill, infants with MMC produced more steps when the context was modified to enhanced sensory input compared to the baseline condition of a typical smooth black belt. Generally, friction and visual flow elicited a greater number of steps than other conditions, producing increases of 47% and 20% respectively, compared to baseline. We observed significant individual variability as well. Future studies should investigate the effect of enhancing sensory input by combining friction and visual flow and with opportunities to practice, thus allowing infants to adapt and, perhaps, settle into more stable response patterns.

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Figure 1.
Scatterplot of infants' raw Bayley Sensorimotor subscale scores, by lesion group and age.

Figure 2. Scatterplot of infants' step rate collapsed over context, by lesion groups, as a function of Ponderal Index.

Figure 3.
Scatterplot of infants' leg activity rate as a function of the Ponderal Index.

Figure 4. Mean percent of total steps that were of each step type, collapsed over conditions, for (A) younger and (B) older age groups. AL= alternating, SI= single, PA- parallel, DO=double.

Figure 5. Steps rate by condition, age and lesion level, (A) low-level lesion, (B) middle-level lesion and (C) high-level lesion.

Figure 6.

Z score for step rate for (A) younger group, (B) older group, (C) low-level lesion, (D) middle-level lesion and (E) high-level lesion. B=baseline, VIS=visual flow, U=unloading, W=weights, VEL=Velcro, F=friction.

Figure 7.
Normalized cycle duration across condition by age groups.

Figure 8.
 Leg activity rate across condition by age groups for (A) low-level lesion, (B) middle-level lesion and (C) high-level lesion.

Table 1

Medical and Anthropometric Characteristics of Participants.

Participant Number	Age (days) at testing	Ponderal Index	Bayley Raw Motor Score	Lesion Level	Hydrocephalus	Shunt	Arnold Chiari II Malformation	Foot deformity	Hip Status
1	64	0.031	18	L5-S1	Y	Y			
2	115	0.023	15	L2	Y	Y	Y		
3	117	0.21	21	S2-S4	Y	Y	Y		
4	120	0.029	23	L5-S1	Y	Y	Y		
5	123	0.028	17	S1					
6	124	0.026	26	L1	Y			R club foot	
7	128	0.034		L2	Y	Y	Y	Bilateral club feet	
8	133	0.026	16	L5	Y	Y		Bilateral club feet	Bilateral - subluxating
9	134	0.027	28	S2					
10	145	0.034	18	L5-S1	Y	Y	Y		R dislocated
11	155	0.03	24	L2-L3	Y	Y	Y	Bilateral club feet	R dysplasia L dislocated
12	231	0.048	10	L5-S2	Y	Y	Y	Bilateral dorsiflexed	
13	249	0.03	49	L4-S1	Y	Y	Y	Bilateral club feet	
14	251	0.021	38	S2-S4	Y			Bilateral dorsiflexed	
15	259	0.032	44	L4-S1					
16	266	0.029	37	L3-L4	Y	Y			
17	277	0.022	50	S3-S4	Y		Y		
18	278	0.037	44	L4-L5	Y		Y	Bilateral club feet	Bilateral - dysplasia R unstable
19	278	0.032	30	L5-S1	Y	Y	Y	Bilateral club feet	
20	281	0.029	33	L4-L5	Y	Y			
21	282	0.027	26	S1	Y	Y		Bilateral dorsiflexed	
22	285	0.027	51	L4-L5	Y		Y		
23	286	0.027	38	L2				Bilateral club feet	
24	303	0.024	53	S1-S4	Y	Y			
25	305	0.027	48	L3-L4	Y	Y	Y	Bilateral club feet	L hip tendon release

Participant Number	Age (days) at testing	Ponderal Index	Bayley Raw Motor Score	Lesion Level	Hydrocephalus	Shunt	Arnold Chiari II Malformation	Foot deformity	Hip Status
26	315	0.026	22	L2-L3	Y	Y		Bilateral club feet	
27	371	0.027	55	S1-S2	Y		Y	Bilateral club feet	R dislocated

Y=yes, L=left, R=right.