A longitudinal study of motor and cognitive development in infants with congenital idiopathic clubfoot
Abstract

It was the aim of this study to examine the motor and cognitive development of infants with congenital idiopathic clubfoot compared to typically developing infants. We repeatedly tested the gross motor, fine motor and cognitive abilities of 12 infants with clubfoot and 12 typically developing infants at the ages of 4, 6, 9, and 12 months with the Bayley-III Scales. All infants with clubfoot were treated with the Ponseti method, which led to a restriction of normal movements of the lower extremities in the first months of life. They showed a great delay in gross motor development but not in fine motor or cognitive development. However, in the clubfoot group, we found some slight deficits in specific cognitive tasks, including problem solving and spatial memory. Additionally, our results revealed significant correlations between gross and fine motor performance and cognitive performance in the control group but only between fine motor and cognitive performance in infants with clubfoot, indicating that both, fine and gross motor skills, are related to cognitive processes and can mutually replace each other to a certain degree. Further research is needed to gain a deeper understanding of clubfoot infants’ development and to clarify the need for mobility training.

Keywords: congenital talipes equinovarus, Ponseti method, cognitive development, motor development, infancy
Introduction

Piaget (1952) presented the argument that children’s cognition originates from their sensorimotor actions. Additionally, Gibson (1966, 1979) emphasized the significance of motor actions in gaining new, meaningful information about objects. Most studies investigated and found evidence of such a link between motor and cognitive development in typically developing infants (for example Clearfield, 2004; Kermoian & Campos, 1988; Schwarzer, Freitag, Buckel, & Lofruthe, 2013). Few studies have tested the consequences of this link in infants with delayed motor development and provided mixed results (for example Campos, Anderson, & Telzrow, 2009; Rivière & Lécuyer, 2002, 2003). While studies of children with locomotor delay due to spina bifida revealed a strong link between motor and cognitive development, indicating cognitive impairments in these children (e.g., Campos, Anderson, & Telzrow, 2009; Lomax-Bream, Taylor, Landry, Barnes, Fletcher, & Swank, 2007; Lehmann & Jansen, 2013), studies of children with locomotor delay due to spinal muscular atrophy did not show any cognitive impairments (Rivière & Lécuyer, 2002, 2003). One possible reason for the conflicting results could be that children with spina bifida and children with spinal muscular atrophy tested in the different studies likely suffered not only from motor impairments but also from other impairments such as neurological problems or poor well-being to different extents. These additional impairments could have caused the varying cognitive results in the different groups. Therefore, it is the aim of the present study to further investigate the link between motor and cognitive development in infants with treated congenital idiopathic clubfoot who only suffer from certain locomotor impairment without any other problems.
Several studies of typically developing infants have emphasized the importance of motor experiences, especially locomotor experiences, in many psychological processes.

First, the development of locomotion has been shown to be related to specific perceptual abilities. Campos, Bertenthal, and Kermoian (1992) tested 7-month-old crawlers and pre-crawlers using the “visual cliff paradigm”. In this experiment, the crawlers but not the pre-crawlers without any walking experience showed wariness of heights as evidenced by significant heart rate changes. Furthermore, Uchiyama and colleagues (2008) demonstrated that locomotor experience is also important for visual proprioception, the sense of self-motion produced by patterns of optic flow (Gibson, 1979). In one study, pre-locomotor infants received self-produced locomotor training via a powered mobility device, which caused a change in their postural compensation and emotional expression to peripheral optic flow in a moving room apparatus (Uchiyama et al., 2008).

Second, locomotor experience has also been found to be relevant to spatial cognition and spatial language. Several researchers have mentioned that infants with enhanced locomotor experiences performed better in spatial cognitive search tasks (Kermoian & Campos, 1988; Bai & Bertenthal, 1992; Clearfield, 2004; Berger, 2010). Furthermore, using a habituation task, Soska, Adolph, and Johnson (2010) showed that the attainment of a self-sitting posture and coordinated visual-manual object exploration skills is related to the ability to complete non-visible parts of three-dimensional objects, as infants’ ability to sit seems to facilitate their visual inspection of objects (Soska et al., 2010). In a longitudinal study of 9- to 36-month-old infants, Oudgenoeg-Paz, Leseman, and Volman (2015) examined the attainment of motor milestones, spatial-relational object exploration and exploration through self-locomotion at 20 months, spatial memory at 24 months, spatial processing at 32 months and spatial language at 36
months. They found that unsupported sitting and independent walking at a younger age are related to later advances in spatial cognition and spatial language. Additionally, they revealed that exploration through self-locomotion partially mediated the relations between attainment of independent walking and spatial language and cognition (Oudgenoeg-Paz et al., 2015). Moreover, Schwarzer and colleagues demonstrated that crawling experience and manual object exploration are related to the mental rotation ability of 9-month-old infants (Schwarzer et al., 2013; Schwarzer, Freitag, & Schum, 2013). Frick and Möhring (2013) verified these results. In one study, they showed that the mental rotation ability of 6-month-old infants improves as a result of their experience with manually exploring objects. In another investigation, the same authors found that 8- and 10-month-old children who were able to walk with assistance performed better in a mental rotation task than infants at the same age with no walking experience (Frick & Möhring, 2013).

Third, self-produced locomotion also has been shown to be important for the development of specific socio-emotional skills. Campos, Kermoian, Witherington, Chen, and Dong (1997) studied referential gestural communication in 8 1/2-month-old infants. They divided the infants into 3 groups: hand and knee crawlers who had been crawling for 6 weeks, pre-locomotor infants and prelocomotor infants with 40 or more hours of experience with moving in a walker. The locomotor and the pre-locomotor children with walker experience were significantly more able to follow the point and gaze of the experimenter correctly than the infants without any locomotor experience.

In sum, recent studies have shown a strong link between locomotion experiences and a variety of psychological processes in typically developing infants. However, these studies did not address the cognitive development of motorically delayed infants.
Influence of motor experiences on cognitive processes in motorically delayed children

Few studies have tested the consequences of the link between motor and cognitive development in locomotor-delayed children. These few studies primarily focused on two specific diseases: spina bifida and spinal muscular atrophy.

Spina bifida is a congenital neural tube defect with varying brain and spine dysmorphias. Therefore, spina bifida patients have difficulties in a variety of domains, including motor difficulties (Lomax-Bream et al., 2007). Campos and colleagues (2009) showed that infants with locomotor delay due to myelomeningocele also experienced delays in specific spatial cognitive and socio-emotional skills. In this study, infants’ performance on an object permanence task and a referential gesture task was poor until they started to crawl. Rivera, Radtka, Anderson and Campos (2012) investigated the spatial cognitive development of infants with myelomeningocele compared to that of typically developing infants in a longitudinal study. Consistent with the findings of Campos and colleagues (2009), infants with spina bifida improved after the onset of crawling ($M = 19.6$ months) in the areas of joint visual attention, shape perception and visual control of posture. Furthermore, in a longitudinal study, Lomax-Bream and colleagues (2007) confirmed the importance of early motor development in different cognitive processes in 6- to 36-month-old infants with spina bifida as well as healthy control infants. In this study, infants with spina bifida showed persistent impaired motor, cognitive, language and daily living skills. In addition, higher-quality parenting had a positive effect on cognitive and language development in both groups. This means that infants of mothers who accurately interpreted and sensitively responded to their infants’ social signs had higher levels and faster rates of growth in the language and cognitive domains (Lomax-Bream et al., 2007). Moreover, a case report by Lynch, Ryu, Agrawal, and Galloway (2009) demonstrated that power mobility training for a 7-month-old boy with spina bifida was helpful in improving the infant’s Bayley-III cognition and language scores. The Bayley Scales were administered just
before the training started when the infant was 7 months of age as well as after the training ended when the boy was 12 months of age. Initially, the infant achieved scores that indicated slight delay in all domains. After the training, at 12 months of age, the infant’s scores for expressive language and fine motor development matched his chronological age. Remarkably, his cognitive and receptive language scores exceeded his chronological age, indicating the supportive role of power mobility training, even if the results were generated from one child (Lynch et al., 2009). Studies with older children with spina bifida showed additional deficits in other domains such as route knowledge (Wiedenbauer & Jansen-Osmann, 2006), math abilities (Barnes, Wilkinson, Khemani, Boudesqui, & Fletcher, 2006), integration of information across sentences (Barnes, Huber, Johnson, & Dennis, 2007) and mental rotation performance (Wiedenbauer & Jansen-Osmann, 2007). In one study by Lehmann and Jansen (2013), 8 to 12 years old children who were restricted in their ability to walk due to spina bifida and hydrocephalus showed an impaired mental rotation performance in comparison to same-aged walking children with hydrocephalus without spina bifida. These results indicated that mental rotation deficits of spina bifida children at that age are most of all due to their locomotor impairment and not due to other neurological deficits (Lehmann & Jansen, 2013). Additionally, another study has shown that juggling training could improve the mental rotation process of 8 to 12 years old children with spina bifida (Lehmann & Jansen, 2012), indicating again the impact of motor experience on the mental rotation ability of children with spina bifida.

Spinal muscular atrophy (SMA) is a genetic neuromuscular disease whereby the alpha motor neurons degeneration lead to muscle weakness without cognitive impairment (Borell, Pechmann, & Kirschler, 2015). SMA cases are divided into different subtypes, and previous studies only focused on type 2. Infants with type 2 SMA have markedly delayed motor development after the attainment of sitting (Borell et al., 2015). Rivière and Lécuyer (2002) demonstrated that 21- to 36-month-old children who were unable to crawl or walk because of
type 2 SMA did not show any delay in a memory-for-location task (Rivière & Lecuyer, 2002).
In another three-location-search task (Rivière & Lécuyer, 2003), the performance of the
children with type 2 SMA was even better than that of the healthy control children. The authors
proposed that the healthy children had problems with inhibiting their direct-finding response,
whereas the type 2 SMA children had more time for inhibition because they were slower to
start their reaching movement (Rivière & Lécuyer, 2003).

Taken together, studies of locomotor-delayed children have yielded conflicting results
concerning the influence of locomotor experiences on cognitive processes. While children with
spina bifida showed strong motor and cognitive impairments (e.g., Lomax-Bream et al., 2007),
children with type 2 SMA, who were not able to crawl or to walk at all did not show any spatial
cognitive impairments (e.g., Rivière & Lécuyer, 2002, 2003).

Theoretical background relating to clubfoot

Congenital idiopathic clubfoot, also called congenital talipes equinovarus, is a foot
deformity in otherwise healthy born infants (Morcuende, Dolan, Dietz, & Ponseti, 2004). In
Central Europe and North America, this deformity with multifactorial causes has a prevalence
of 1-2 in 1000 newborns. For fifty percent of them, both feet are affected, and it is more common
in boys than in girls (Kujus, 2005; Westhoff, Weimann-Stahlschmidt, & Krauspe, 2008).
Typically, as seen in Figure 1, the foot is displaced and rotated (Ponseti & Smoley, 2009) and,
therefore, has to be treated soon after birth. The Ponseti method is a conservative and
nonsurgical treatment for idiopathic clubfoot with good functional results, fewer operative
interventions and a relapse rate that is comparable to that of other treatments (for example
Eberhardt, Peterlein, Fernandez, & Wirth, 2012; Halanski, Davison, Huang, Walker, Walsh, &
Crawford, 2010; Laaveg & Ponseti 1980; Morcuende et al., 2004). It includes weekly
manipulation and casting (see Figure 2) by an orthopedic specialist and, if needed, a
percutaneous Achilles tenotomy and foot abduction bracing (Krauspe, Westhoff, & Wild, 2006). To avoid relapse, patients have to wear the foot abduction orthosis (see Figure 3) full time for three months, then for another two to four years only while sleeping (Ponseti & Smoley, 2009; Radler, 2013; Westhoff et al., 2008). Thus, the Ponseti method causes a restriction of normal movements of the lower extremities in the first months of life.

Until now, only a few studies have focused on the motor development of infants with treated idiopathic clubfoot. For example, Sala, Chu, Lehman, and van Bosse (2013) interviewed the parents of 36 infants with Ponseti-treated idiopathic clubfoot repeatedly concerning the achievement of eight gross motor milestones. The parents reported minimal delays of 0.7 to 2 months in achieving the following six gross motor milestones: “roll prone to supine”, “sit without support”, “crawling on hands and knees”, “pull to stand”, “cruising” and “independent ambulation”. Furthermore, Masquijo, Campos, Torres-Gomez, and Allende (2013) studied the locomotor development of healthy infants and infants with either idiopathic clubfoot or developmental dysplasia of the hip. The authors interviewed the parents regarding the age at which the children could sit without support, crawl and walk. In comparison to the healthy infants, the other two groups showed a non-significant slight delay in the age at which they began to crawl and a significant delay in the onset of walking. Another study examined infants with treated clubfoot and children without any diagnosis at the ages of 3, 6, 9 and 12 months using the Alberta Infant Motor Scale (Garcia, McMulkin, Tompkins, Caskey, Mader, & Baird, 2011). The results suggested that the clubfoot group achieved significantly lower scores at 9 and 12 months but not at 3 and 6 months. Finally, Zionts, Packer, Cooper, Ebramzadeh, and Sangiorgio (2014) focused on the onset of independent walking in infants with Ponseti-treated idiopathic clubfoot. They interviewed their parents and observed the infants’ ability to walk independently. The authors determined that the children with idiopathic clubfoot achieved independent walking approximately two month later than normally developing children.
Moreover, walking age was also affected by the severity of the deformity and the occurrence of a relapse (Zionts et al., 2014).

In sum, previous studies suggested a slight delay in the gross motor development of infants with clubfoot, especially in the second part of the first year of life, particularly in crawling and independent walking (Garcia et al. 2011; Masquijo et al., 2013; Sala et al., 2013; Zionts et al., 2014). However, previous studies do not explain whether the cognitive development of infants with congenital idiopathic clubfoot is delayed. Thus, continued work on this topic is necessary because it is conceivable that delayed locomotor development due to clubfoot is a key risk factor for cognitive impairment. This would highlight the need for mobility training for all infants with congenital clubfoot or other locomotor disabilities. Additionally, it is important to investigate fine motor abilities in clubfoot infants, because several studies have shown that fine motor skills are related to cognitive processes, too (e.g. Schwarzer et al., 2013; Soska et al., 2010). For example, Schwarzer and colleagues (2013) have demonstrated that 9-months old non-crawlers who spontaneously explored objects in an advanced manner, performed better in a mental rotation task in comparison to 9-months-old non-crawlers who spontaneously explored objects less.

**The current study**

To investigate the link between locomotor experience and cognitive abilities in infants with delayed motor development, we compared infants with clubfoot to healthy control infants. This comparison allows us to examine this link when the groups only differ in the extent of their locomotor experience without any other cognitive or neural problems. We repeatedly measured the motor and cognitive development of infants with treated idiopathic clubfoot and normally developing infants using the Bayley Scales of Infant Development III (Bayley, 2006). The Bayley-III Scales measures the general development of infants across different domains
but several items also provide information about the infants’ development in certain motor and
cognitive aspects. Therefore, the analyses of the Bayley data allowed us to study the association
between motor and cognitive development in general and examine the relation between certain
locomotor abilities and cognitive abilities such as spatial cognitive abilities. Based on previous
studies (Garcia et al. 2011; Masquijo et al., 2013; Sala et al., 2013; Zionts et al., 2014), we
expected that infants with clubfoot would be delayed in gross motor development but we did
not expect to find any impact on fine motor development. Regarding cognitive development
and the relationship between motor and cognitive development, we had no definite predictions.
On the one hand, following the work on infants with spina bifida (Campos et al., 2009; Lomax-
Bream et al., 2007; Rivera et al., 2012), it could be that infants with congenital idiopathic
clubfoot would be delayed in their cognitive development, especially in their spatial cognitive
development. On the other hand, given the studies of children with locomotor impairment due
to spinal muscular atrophy (Rivière & Lécuyer, 2002, 2003), it could be that infants with
locomotor delay due to clubfoot would not show any cognitive difficulties.

**Method**

**Ethics statement**

The present study was conducted in full accordance with the Research Ethics Guidelines
of the German Psychological Society (DGPs). Prior to the first test, written consent to
participate in the study was obtained from the infants’ parents. The Office of Research Ethics
at the University of Giessen approved the experimental procedure and the informed consent
protocol.

**Participants**

A total of 24 infants participated in this study, 12 infants (1 female, 11 males) with
congenital idiopathic clubfoot and 12 typically developing infants (1 female, 11 males) without
any diagnosis. Two additional infants with congenital idiopathic clubfoot were excluded from the study because they were born prior to the 37th week of pregnancy. We excluded the two corresponding control children as well. Thus, all 24 children were born at term. The infants with congenital clubfoot had no other major disabilities and were recruited from the orthopedic clinic at the university hospital in [redacted], where they were treated with the Ponseti method. The treatment of all infants with congenital idiopathic clubfoot was the same and started within the first two months after birth. Four infants had two deformed feet and eight infants had only one deformed foot. The typically developing infants were recruited from a local hospital in [redacted] where they were born. We matched the infants in the two groups on gender and age. The control infants were assessed at the same age as the clubfoot infants +/- 3 days. That is, we tested a healthy control infant within three days of testing the corresponding clubfoot child. In the majority of cases, the participating infants were Caucasian, middle to upper-middle class and living in urban and suburban areas of [redacted].

Measures

The infants’ parents completed a demographic questionnaire with items relating to their education, family status and age at birth. This questionnaire was also used to assess the infants’ weight at birth, Apgar values and number of siblings. We assessed infants’ motor and cognitive development using the German version of the Bayley Scales of Infant Development III (Bayley, 2006). The cognitive subscale of the Bayley-III measures sensorimotor development, exploration and manipulation, object relatedness, concept formation, memory and other aspects of cognitive processing (Bayley, 2006). The motor subscale includes a fine motor subtest and a gross motor subtest. The fine motor scale items assess prehension, perceptual-motor integration, motor planning and motor speed, whereas the gross motor scale items measure static positioning, dynamic movement, balance and motor planning (Bayley, 2006). All three subscales are based on direct child interaction, and the experimenter rates the performance of
the child immediately during testing. Each subtest of the Bayley-III involves different sets of items, and the sum of the scores of passed items provides information about the infant’s developmental status. The Bayley-III provides different types of norm-referenced scores from the subtests’ total raw scores. Bayley scaled scores range from 1 to 19, with a mean of 10 and a standard deviation of 3 and represent a child’s performance relative to same-age peers (Bayley, 2006).

**Procedure**

The experimenter was trained in the administration and interpretation of the Bayley-III in advance. All infants were tested at 4, 6, 9 and 12 months of age individually at home in their familiar environment. Each testing session took approximately 1 to 1.5 hours. When testing ended, each infant received a present and a certificate of participation. Prior to the first testing session, the parents completed the demographic questionnaire and the consent form. All tests were videotaped, and the order of the three different Bayley subscales was randomized. According to the manual (Bayley, 2006), each subscale test started with an appropriate start item that corresponded with the chronological age of the infant and continued until the infant failed five consecutive items. If an infant failed one of the first three items, the items corresponding with the previous age group were presented.

**Data analysis**

We used SPSS 22.0 for all statistical analyses. To analyze the sample characteristics, we conducted independent samples t-tests. Moreover, we conducted three different linear mixed models with group and time of measurement as fixed factors to analyze the gross motor, fine motor and cognitive development of both groups longitudinally. Each time, we chose a first-order linear autoregressive AR (1) model, which allowed us to include missing and correlated data and therefore minimize the loss of data associated with repeated measurements designs. A
large value of the correlation parameter rho ($\rho$) and a significant $p$-value of the Wald test indicate that the autoregressive structure of the model fits the data (Bühl, 2012). Furthermore, we compared the performance of both groups regarding individual cognitive items with chi-squared tests. Finally, we conducted bivariate correlations between the motor and cognitive scaled scores.

**Results**

**Sample characteristics**

Table 1 shows the sociodemographic, birth weight and Apgar data. The results of the t-tests did not show any significant differences between the two groups.

**Gross motor development**

A first-order linear autoregressive AR (1) model (AR1, $\rho = 0.520, p < .000$) with the Bayley gross motor scaled score as the dependent variable and group (between subjects: clubfoot, control) and time of measurement (within subjects: 4 months, 6 months, 9 months, 12 months) as fixed factors revealed a significant main effect of group, $F(1, 27) = 28.32, p < .001$, $d = 1.51$. Moreover, the linear mixed model revealed a main effect of time of measurement, $F(3, 59) = 4.10, p = .010$, and a marginal significant interaction between time of measurement and group, $F(3, 59) = 2.72, p = .053$. As seen in Figure 4, results of the Bonferroni-corrected post hoc t-tests were significant at 6, 9 and 12 months of age. Thus, at 6, 9 and 12 months of age, infants with congenital clubfoot showed significantly lower gross motor performance than the healthy control infants. Furthermore, their gross motor scaled scores decreased across time, whereas the scores of the healthy control infants remained stable.

**Fine Motor Development**
A first-order linear autoregressive AR (1) model (AR1, ρ = 0.56, p < .001) with the Bayley fine motor scaled score as the dependent variable and group (between subjects: clubfoot, control) and time of measurement (within subjects: 4, 6, 9, and 12 months) as fixed factors did not reveal any differences between the clubfoot and the control infants regarding their fine motor performance, $F(1, 23) = 2.22$, $p = .150$, $d = 0.52$. Thus, fine motor performance of both groups was similar. Surprisingly, we found a significant main effect of time of measurement, $F(3, 54) = 5.80$, $p = .002$. As seen from Figure 5, across time the Bayley fine motor scaled scores increased in both groups. Furthermore, the model did not reveal an interaction effect between time of measurement and group, $F(3, 54) = 0.66$, $p = .583$.

**Cognitive Development**

We conducted a first-order linear autoregressive AR (1) model (AR1, ρ = 0.54, p < .001) with the Bayley cognitive scaled score as the dependent variable and group (between subjects: clubfoot vs. control) and time of measurement (within subjects: 4, 6, 9, and 12 months) as fixed factors. As seen in Figure 6, the model did not reveal a significant main effect of group, $F(1, 22) = 3.08$, $p = .090$, $d = 0.66$, a significant main effect of time of measurement, $F(3, 54) = 1.98$, $p = .129$, or a significant interaction between group and time of measurement, $F(3, 54) = 0.57$, $p = .641$. Thus, comparable to the healthy control infants, infants with congenital clubfoot did not show significantly lower cognitive performance per se.

We further compared the cognitive performance of both groups on each cognitive item of the Bayley Scales individually with chi-squared tests to uncover whether group differences existed in certain cognitive tasks. The results showed that the clubfoot infants had significantly lower performance only on two different cognitive tasks (see Figure 7a and 7b). At 6 months of age, they were less able to pull a cloth purposely toward them to obtain an object placed on the edge of the cloth (Item 28), $\chi^2(1, N = 20) = 5.45$, $p = .04$, $d = 1.22$. Additionally, at 12
Clubfoot infants' motor and cognitive development

months of age, clubfoot infants performed more poorly on a spatial cognitive search task. Specifically, they were significantly less able to find a hidden object by looking under the correct one of two washcloths (Item 40), \( \chi^2 (1, N = 23) = 6.14, p = .030, d = 1.21 \). On all other cognitive items, the two groups achieved comparable results.

**Correlations between motor and cognitive Bayley scores**

Table 2 shows the correlations between gross motor and cognitive scaled scores. In the control group the gross motor scaled score was significantly correlated with the cognitive scaled score with 4 months \( (r_s = .798, p = .003) \) and 12 months \( (r_s = .744, p = .006) \) of age. In the clubfoot group none of these correlations were significant (see Table 2).

Table 3 presents the correlations between fine motor and cognitive scaled scores. In the control group the fine motor scaled score was significantly correlated with the cognitive scaled score with 4 months \( (r_s = .668, p = .035) \) and 12 months \( (r_s = .770, p = .003) \) of age. In the clubfoot group fine motor performance was significantly correlated with cognitive performance with 6 months \( (r_s = .782, p = .007) \), 9 months \( (r_s = .690, p = .013) \) and 12 months \( (r_s = .675, p = .023) \) of age (see Table 3).

**Discussion**

The aim of the current study was to investigate the relation between motor and cognitive development in infants with congenital idiopathic clubfoot who suffer only from locomotor impairment without any other problems.

The current study yielded several important results and implications. First, we verified gross motor delays in infants with Ponseti-treated idiopathic clubfoot. We found obvious gross motor delays at 6, 9 and 12 months of age. Moreover, the gross motor scaled scores of the clubfoot children decreased across the age groups. Figure 4 illustrates the increasing discrepancy between clubfoot infants and typically developing infants gross motor
performance. Previous studies (Garcia et al., 2011; Masquito et al., 2013; Sala et al., 2013; Zionts et al., 2014) demonstrated only minimal gross motor delays in infants with clubfoot, especially in the second half of the first year of life, particularly in crawling and independent walking. However, in contrast to our work, most previous studies simply involved interviews of the parents regarding the motor development of their infants and can, therefore, be seen as limited in terms of the accuracy of the parents’ reports (Masquito et al., 2013; Sala et al., 2013; Zionts et al., 2014). Furthermore, some of the previous studies did not test a group of healthy control infants. They simply compared the values of the clubfoot infants with published values for typically developing infants (Sala et al., 2013; Zionts et al., 2014). Hence, our study indicates that the manipulation, casting and bracing of congenital idiopathic clubfoot might have a stronger impact on infants’ gross motor development than expected.

Second, the present results revealed that infants with congenital idiopathic clubfoot showed age-appropriate fine motor performances. Surprisingly, the fine motor performance of the clubfoot infants and the control infants was the lowest at 4 months of age and increased with age. The low performance of the 4-month-old clubfoot and control infants might be explained by the fact that, in general, 4-month-old boys have suspected fine motor developmental delays significantly more often than girls, as revealed in the study by Valla, Wentzel-Larsen, Hofoss, and Slinning (2015). In our study, 22 boys and only 2 girls participated. It could be that the generally lower values of 4-month-old boys caused the low fine motor values in our study compared to the US Bayley norms, which rely on an equal number of female and male infants. Nevertheless, the fine motor scaled scores of all age groups were within the normal range of two standard deviations below and above the mean of 10.

Third and most importantly, we did not find any general cognitive impairments in infants with congenital idiopathic clubfoot. Although their gross motor scaled scores dramatically decreased with age, their cognitive scaled scores remained stable and were average at each
measurement time point and not significantly different from those of the control group. Consequently, our results did not confirm the findings of Lomax-Bream and colleagues (2007), who reported persistent cognitive impairments in infants with locomotor delay due to spina bifida. However, infants with spina bifida are not comparable to infants with clubfoot because they can have spine and brain dysmorphias (Lomax-Bream et al., 2007). Therefore, in addition to their locomotor deficits, they can experience numerous difficulties in a variety of other domains (e.g., Wiedenbauer & Jansen-Osmann, 2006; Barnes et al., 2006, 2007). Although the clubfoot infants in our study did not show any general cognitive impairments, they performed significantly poorer on two specific cognitive tasks: a problem-solving task at 6 months of age and a spatial cognitive search task at 12 months of age. In these tasks, the scores of the clubfoot infants were clearly lower than those of the control infants. One explanation for the observed delays could be that locomotor-delayed infants are less able to reach and explore their environment in a self-induced manner and, therefore, are trained to a lesser extent in facing specific problems, such as recovering objects. Furthermore, pre-locomotor infants are generally less attentive during spatial cognitive search tasks (Campos et al., 2000) and, consequently, perform more poorly on these tasks. Thus, our results suggest that, beyond normal general cognitive development, there seems to be a certain kind of impairment in specific cognitive tasks, including problem solving and spatial cognition, that parallels the corresponding link in healthy infants (Bai & Bertenthal, 1992; Berger, 2010; Clearfield, 2004; Kermoian & Campos, 1988) and infants with locomotor delay due to spina bifida (Campos et al., 2009). Consequently, our results did not confirm the findings of Rivière and Lécuyer (2002, 2003), who reported that children with spinal muscular atrophy, who were unable to crawl or to walk at all, did not show any delay in a spatial cognitive search task.

Finally, we found significant correlations between gross motor and cognitive developmental scores only in the control group at the ages of 4 and 12 months. In the group of
Clubfoot infants’ motor and cognitive development

infants with locomotor delay due to congenital idiopathic clubfoot we did not find any significant correlation between gross motor and cognitive developmental scores. However, we found significant correlations between fine motor and cognitive developmental scores at the ages of 4 and 12 months in the group of typically developing infants and at the ages of 6, 9 and 12 months in the group of locomotor delayed infants due to congenital idiopathic clubfoot. Our findings confirm and extend previous results on the influence of fine motor experiences on cognitive processes in typically developing infants (e.g. Schwarzer et al., 2013; Soska et al., 2010) and show that both, fine motor and gross motor experiences are related to cognitive performance. These results give rise to the question whether fine motor skills could compensate for impaired gross motor abilities to a certain extent. We speculate that such a compensation may be possible but is has to be considered that clubfoot infants in our study performed clearly poorer on two specific cognitive tasks: a problem-solving task at 6 months of age and a spatial cognitive search task at 12 months of age. Thus, it seems that only locomotor experiences significantly contribute to some specific cognitive processes, including problem solving and spatial memory and could probably not be compensated by fine motor experiences.

Limitations and future directions

The main limitation of the current study is the small sample size. We excluded all infants who were born prior to the 37th week of pregnancy or who had other major disabilities. Therefore, only 24 infants, 12 typically developing infants and 12 infants born with congenital idiopathic clubfoot, participated in this study. Despite the small sample sizes the analyses revealed significant differences between the samples with large effect sizes (Cohen, 1988) indicating considerable differences between the infants with clubfoot and control infants tested. Nevertheless, future studies should investigate the motor and cognitive development of infants with congenital idiopathic clubfoot with a larger sample to verify our results. Furthermore, future research is needed to examine the motor and cognitive development of infants with
congenital idiopathic clubfoot over a longer period of time to clarify whether their gross motor delay and their deficits in spatial memory and problem-solving tasks persist through toddlerhood and childhood. If so, an early motor intervention program might be essential to avoiding persistent difficulties. Moreover, previous studies with healthy infants and infants with locomotor delay due to spina bifida have emphasized the importance of motor experiences for referential gesture communication (e.g. Campos et al., 1997, 2009) and mental rotation (e.g. Schwarzer et al., 2013; Lehmann & Jansen, 2013), too. Thus, a mental rotation task and a following of point and gaze gesture task with locomotor delayed infants due to congenital idiopathic clubfoot could verify the importance of self-produced locomotion in referential gesture communication and mental rotation processes. Additionally, it would be of interest to study the associations between gross motor, fine motor and cognitive abilities in infants with either fine motor or gross motor delay (clubfoot). If fine motor and gross motor experiences are related to cognitive performance and could mutually replace each other, both groups should not have any general cognitive impairments.

Despite the small sample size in this study, our results indicated that infants with congenital idiopathic clubfoot are markedly delayed in gross motor development. Pediatric orthopedists should communicate these findings to parents to avoid unrealistic expectations regarding their infants’ achievement of gross motor milestones. The general cognitive performance of clubfoot infants was comparable to that of the healthy control infants, except for two specific cognitive tasks requiring problem solving and spatial memory. Hence, it seems that motor development, especially locomotion, does not have a strong impact on cognitive development per se, but it seems to facilitate the development of specific cognitive skills.
Author Contributions

All authors developed the concept. JD conducted the study and wrote the first draft of the manuscript. All authors contributed to and have approved the final manuscript.

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Conflict of Interest Statement

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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References


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Barnes, M. A., Wilkinson, M., Khemani, E., Boudesquie, A., Dennis, M., & Fletcher, J. M.


Table 1

Sample characteristics

<table>
<thead>
<tr>
<th></th>
<th>Clubfoot group</th>
<th>Control group</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>( n = 12 )</td>
<td>( n = 12 )</td>
</tr>
<tr>
<td>Birth weight (kg)</td>
<td>3.4</td>
<td>3.7</td>
</tr>
<tr>
<td></td>
<td>0.4</td>
<td>0.6</td>
</tr>
<tr>
<td>Apgar (1 min)</td>
<td>9.4</td>
<td>9.1</td>
</tr>
<tr>
<td></td>
<td>0.7</td>
<td>0.3</td>
</tr>
<tr>
<td>Apgar (5 min)</td>
<td>9.9</td>
<td>10.0</td>
</tr>
<tr>
<td></td>
<td>0.3</td>
<td>0.0</td>
</tr>
<tr>
<td>Number of siblings</td>
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<td>0.6</td>
</tr>
<tr>
<td></td>
<td>1.4</td>
<td>0.5</td>
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<tr>
<td>Parental level of education</td>
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<td>3.7</td>
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<tr>
<td></td>
<td>1.2</td>
<td>1.2</td>
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<tr>
<td>Mother’s age (years)</td>
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<tr>
<td></td>
<td>6.4</td>
<td>4.8</td>
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<tr>
<td>Father’s age (years)</td>
<td>33.8</td>
<td>33.7</td>
</tr>
<tr>
<td></td>
<td>6.9</td>
<td>5.9</td>
</tr>
</tbody>
</table>

Note. Parental level of education ranged from 0 = no graduation to 6 = PhD for each parent. The average of the two scores was used in the analyses.
### Table 2

*Correlations between gross motor and cognitive scaled scores*

<table>
<thead>
<tr>
<th></th>
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<tr>
<td></td>
<td>((n = 12))</td>
<td>((n = 12))</td>
</tr>
<tr>
<td></td>
<td>(r_s)</td>
<td>(p)</td>
</tr>
<tr>
<td>4 months</td>
<td>.459</td>
<td>(ns)</td>
</tr>
<tr>
<td>6 months</td>
<td>.153</td>
<td>(ns)</td>
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<tr>
<td>9 months</td>
<td>.255</td>
<td>(ns)</td>
</tr>
<tr>
<td>12 months</td>
<td>-.029</td>
<td>(ns)</td>
</tr>
</tbody>
</table>

### Table 3

*Correlations between fine motor and cognitive scaled scores*

<table>
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<th>Control group</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>((n = 12))</td>
<td>((n = 12))</td>
</tr>
<tr>
<td></td>
<td>(r_s)</td>
<td>(p)</td>
</tr>
<tr>
<td>4 months</td>
<td>.544</td>
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<td>.013</td>
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<tr>
<td>12 months</td>
<td>.675</td>
<td>.023</td>
</tr>
</tbody>
</table>
Figures and Photos

Figure 1. Newborn with congenital idiopathic clubfoot.

Figure 2. Casting of the congenital idiopathic clubfoot.

Figure 3. Infant with clubfoot wearing a foot abduction orthosis.

Figure 4. Mean Bayley gross motor scaled scores and standard errors of both groups at 4, 6, 9 and 12 months of age.

Figure 5. Mean Bayley fine motor scaled scores and standard errors of both groups at 4, 6, 9 and 12 months of age.

Figure 6. Mean Bayley cognitive scaled scores and standard errors of both groups at 4, 6, 9 and 12 months of age.

Figure 7a. Performance of both groups on one specific cognitive task with 6 months of age, 0 = child did not manage the task and 1 = child managed the task.

Figure 7b. Performance of both groups on one specific cognitive task with 12 months of age, 0 = child did not manage the task and 1 = child managed the task.