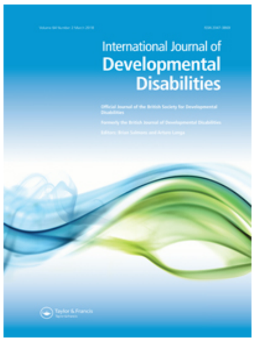

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Associations between tactile localization and motor function in children with motor deficits

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Children with developmental disorders often have poor motor performance. This study aimed to address the association between tactile localization ability, an indicator of body image, and motor function in children with motor deficits. Eighteen children with motor deficits participated, and their upper and lower limbs were assessed. To assess the level to which the patient's body image was developed, a tactile localization task (TLT) was used. In the TLT, experimenters touched a child's fingers, toes, or lower extremities (L/E), and the participants were asked to identify the location of the touch on a body part illustration. We compared TLT ability between high and low motor function groups, and investigated the correlation between TLT and the measures of motor function, age, and non-verbal intelligence. The high motor function group had significantly higher L/E TLT scores than the low motor function group, except in the tests involving the fingers and toes. Furthermore, the L/E TLT correlated only with motor function measures (Gross Motor Function Measure score, measured using one-leg standing time and one-leg hopping ability). The results suggest that children with motor deficits experience developmental delay in terms of their body image.

Keywords: tactile localization, body image, cerebral palsy, developmental disorder, motor deficits

Introduction

Body representation can be measured through various means. Pointing to one's own body parts is a common measure for indicating accurate body representation, and previous studies have often used this method to assess distorted body representation (Cardinali *et al.* 2011; De Vignemont 2010; Paillard 1999; Rossetti *et al.* 1995; Schwoebel and Coslett 2005). Since the seminal neuropsychological work by Head and Holmes (1911–1912), several authors have proposed at least two representations: body schema and body image. Body schema can be defined as a system of preconscious, subpersonal processes that play a dynamic role in governing posture and movement, which serves to guide actions. Body image is often defined as a conscious idea or mental representation of one's own body, and is an intentional content of consciousness that consists of perception, attitudes, and beliefs pertaining to one's own body (Gallagher and Cole 1995). Anema *et al.* (2009) showed double dissociation (body schema and body image) with respect to tactile localization capacities in two stroke patients who retained intact somatosensory function. The authors asked the patients to localize a tactile stimulus by either pointing directly to their stimulated hand or to a pictorial map of the hand. The first task involved the body representation as body schema, whereas the second task assessed the body image. One patient was unable to identify where she had been touched on a line drawing of a hand, but was able to point accurately toward the actual position on her/his hand. The reverse pattern was observed in the other patient. Interestingly, Osumi *et al.* (2015) reported that tactile localization training is important for improving body image in patients with complex regional pain syndrome. De Vignemont (2010) suggested that some forms of body representations are necessary in order to point to the location of touch. In other words, visual equivalence or representation must be established in order for subjects to correctly respond to a request to point to stimulation sites on a drawing or picture of the body parts following tactile stimulation of body parts by others. Patients must also have understood that the drawing represents their own body. Therefore, it is assumed that the ability to identify the perceived location of the tactile stimulus on the illustration of the body parts represents recognition of the body image.

From a developmental perspective, Benton's (1955) classic study reported that normally developing children are able to correctly point to a single touched finger at 6–9 years of age. Additionally, Lefford *et al.* (1974) showed that preschool children could localize where they have been touched with their own fingers before using a map of the hand. This is because localization of the finger on the map of a hand involved a terminus of action that

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was a representation of the body, rather than the subject's own body. For that reason, the authors determined that the ability to understand representation and correspondence of the fingers increases between 3 and 5 years of age. In recent studies, Yoshioka *et al.* (2013) showed that 4-year-old children made large stimulus localization errors on a map of the hand; however, the magnitude of error rapidly decreased with age.

The inability to localize the fingers of one's own hands by pointing is associated with cerebral injuries such as cerebral palsy (CP) (Auld *et al.* 2012; Bhojne and Rege 2001; Boll and Reitan 1972) and other developmental disorders (intellectual disabilities, learning disorder, dyslexia, developmental coordination disorder (DCD), attention deficit/hyperactivity disorder, autism spectrum disorder (ASD)) (Beaton *et al.* 2006; Benton 1955; Elbasan *et al.* 2012; Fletcher *et al.* 1982; Iwanaga *et al.* 2006; Lindgren 1978; Nydén *et al.* 2004; Sotozaki and Parlow 2006). Children with any of the previously mentioned developmental disorders also have motor control dysfunction, known as 'clumsiness' or DCD (Green *et al.* 2002; Miyahara *et al.* 1997; Piek *et al.* 1999; Piek and Dyck 2004; Provost *et al.* 2007). Motor dysfunction in children with developmental disorders is likely due to undeveloped body image. To our knowledge, however, there has been no investigation on the association between their body image and motor function, particularly with a focus on the lower limbs. Understanding the association between body image and motor performance may provide important information for rehabilitation of children with motor deficits. The purpose of this study was to clarify the association between tactile localization performance, as an indicator of body image, and upper and lower limb motor function in children with motor deficits, who had been clinically diagnosed with mild CP, intellectual disabilities (ID), ASD, and attention deficit hyperactivity disorder (ADHD). These children were included because we believe motor impairment in these conditions to be a 'spectrum disorder' on some level. Pearsall-Jones *et al.* (2010) proposed that CP and DCD have similar causal pathways and may fall on a continuum of movement disorder rather than being discrete categories. CP and DCD have some common pre-, peri-, and neo-natal risk factors, indicating potential similarities in etiology, and there is some evidence of similarities in their neural structures (Williams *et al.* 2014).

We hypothesized that, compared to children with high motor function, those with low motor function would have difficulties in recognizing a body part touched by others. Furthermore, we hypothesized that the tactile localization ability would correlate with measures of motor function.

Methods

Participants

Eighteen children with motor deficits participated in this study (12 boys and 6 girls, mean age: 3.4 years, standard deviation [SD] = 1.4), including patients with mild CP caused by periventricular leukomalacia, ADHD, ASD, and ID. Each child underwent a standardized evaluation protocol that included a neurological evaluation and diagnosis by a pediatrician. The Manual Ability Classification System (MACS) and Gross Motor Function Classification System (GMFCS) were used to classify the skill level in the patients' daily lives. These scales are functional, five-level classification systems were used to evaluate motor impairment. Based on the MACS and GMFCS levels, we divided the participants into high and low motor function groups according to both manual and gross motor skills. Their demographic and clinical data are shown in Table 1.

All participants had clumsiness or delayed childhood motor development and were brought to Japan Baptist Hospital for rehabilitation therapy. Inclusion criteria were patients with diagnosed developmental disorders, normal or corrected to normal vision, and comprehension of the basic instructions for performing the measurements. Patients were excluded if they could not complete all the tasks. The participants' parents provided written informed consent prior to testing. The study was conducted in accordance with the Declaration of Helsinki guidelines and was approved by the ethics committee at Kio University.

Materials and procedure

All children completed the following five-tactile localization tasks used to investigate their body images of their upper and lower extremities: the five-finger tactile localization task (5-finger TLT), three-finger tactile localization task (3-finger TLT), three-toe tactile localization task (3-toe TLT), five-toe tactile localization task (5-toe TLT), and lower extremities tactile localization task (L/E TLT). Upper extremity (U/E) motor function was measured using pegboard and touch speed tests. L/E motor function

Table 1 Clinical description of the study participants

Case	Diagnosis	Age (mo)	Sex	GM-FCS	MACS	RCPM
1	ADHD	59	F	I	I	13
2	ASD	53	M	II	I	16
3	PVL	58	F	I	I	21
4	ASD	43	M	II	II	9
5	PVL	119	M	III	II	27
6	PVL	135	M	III	II	22
7	PVL	72	M	III	III	18
8	PVL	101	M	I	II	28
9	PVL	70	F	I	II	14
10	PVL	61	M	I	I	23
11	ID	154	M	I	II	20
12	ID	90	F	I	II	15
13	ID	46	F	II	III	17
14	ASD	93	M	I	I	30
15	ASD	42	M	II	I	16
16	PVL	120	F	III	I	32
17	ASD	92	M	I	I	28
18	PVL	58	M	III	III	19

Abbreviations: ADHD = attention deficit hyperactivity disorder; ASD = autism spectrum disorder; ID = intellectual disabilities; PVL = periventricular leukomalacia; GMFCS = Gross Motor Function Classification System; MACS = Manual Ability Classification System; RCPM = Raven Colored Progressive Matrices; M = male; F = female

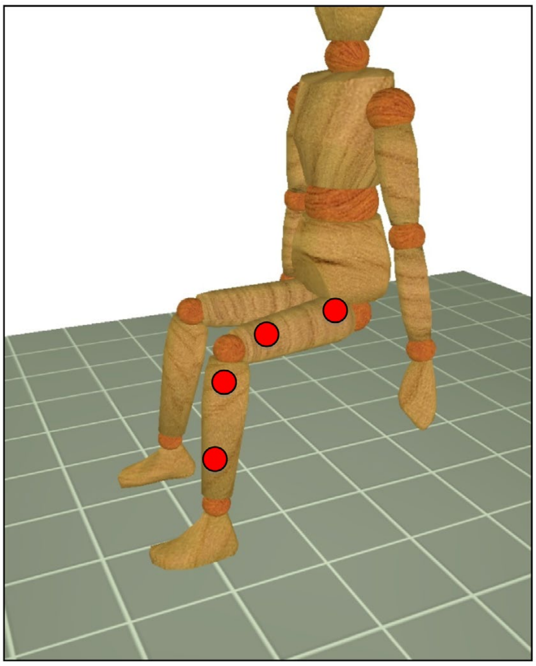

Figure 1 Illustration used in the L/E TLT. In the L/E TLT, the experimenter stimulated one of the four designated areas. Next, participants were asked to point toward the perceived location of the tactile stimulus on the illustration (one of four parts showed by the circles). Abbreviations: TLT = tactile localization task; L/E = lower extremity

was measured via one-leg standing and one-leg hopping tasks and two dimensions (Standing & Walking/Running/Jumping) of the Gross Motor Function Measure (GMFM-88). We also administered Raven's Coloured Progressive Matrices (RCPM) as a non-verbal intelligence test. All assessments were performed within one month by highly experienced pediatric physical therapists.

Tactile localization task

The subjects were seated across from the experimenter and instructed to place their palm down on a table with the fingers spread out. A white board on which hands were drawn was placed between the children and their hands, so that the children's hands could not be seen. The experimenter touched and pressed one of the child's fingernails. The child was then asked to point to the corresponding drawn finger with the index finger of the opposite hand. These procedures were conducted similar to the methods described in previous studies (Gracia-Bafalluy and Noël 2008; Noël 2005). In order to preclude the influence of sensory disturbances, the examiner constantly confirmed if the children were aware of being touched during testing by asking for a simple 'yes' or 'no' answer.

In the 5-finger TLT, the right or left hand was selected at random, and each finger (thumb to little finger) was touched twice, for a total of 10 randomly ordered touches. In the 3-finger TLT, three fingers (thumb, middle, and little) were each touched three times, for a total of nine randomly ordered touches. The 3 and 5-toe TLTs were performed for the toes in a similar fashion. In the L/E TLT, four locations (proximal and distal parts of the thigh, and proximal and distal parts of the lower leg) were touched twice, for a total of eight touches (Figure 1). The total correct response score was converted to a percentage, and data were expressed as the correct answer rate.

Pegboard test

The Purdue Pegboard test was used as a measure of dexterity, and was performed with vision allowed. Children were required to insert small pins into holes on the board using only the right or left hand. The examiner timed them for 30 s and then counted the number of pins successfully placed in the holes. Previous studies have shown that one-trial administration of the Purdue Pegboard is a sufficiently reliable assessment (Amirjani *et al.* 2011; Gallus and Mathiowetz 2003; Wilson *et al.* 1982). This test has been used in studies comparing timed motor performance between children with DCD and control children (Cantell *et al.* 1994; Pitcher *et al.* 2003).

Touch speed test

Application software for the Apple iPad (In-Trinity Co., Ltd) was used to evaluate hand reaction time in response to visual cues. After observing an experimenter's iPad (Apple, iPad2, USA) demonstration, children underwent a training session to familiarize themselves with the test procedure. During the actual testing procedure, they were asked to touch a component of a 12-part schematic, illuminated in red, in rapid succession with either the right or left index fingers. The mean response time and number of errors due to touch errors in five consecutive trials were calculated.

One-leg standing and one-leg hopping tasks

The ability to maintain a one-leg standing position and hop on one foot without postural support was also assessed. We measured standing time on one leg with a digital stopwatch and used the longest time out of five trials for data analysis. The maximum value for the measurement was 20 s. In the hopping task, we used the maximum number of hops achieved out of 10 trials. The maximum value was 20 hops. These tasks were carried out on gym mattresses in order to prevent injuries.

GMFM-88

The GMFM-88 is a clinical tool designed to evaluate changes in gross motor function in children with CP (Russell *et al.* 2002). Items on the GMFM-88 span the spectrum from activities like lying and rolling up to walking, running, and jumping skills. While the original measure was designed and validated for children with CP, there is evidence that the GMFM-88 is also valid for use in children with Down syndrome (Gémus *et al.* 2001; Palisano *et al.* 2001; Russell *et al.* 1998). Accordingly, we used two dimensions (Standing and Walking/Running/

Jumping) of the GMFM-88 in order to assess static and dynamic standing balances.

RCPM

The RCPM (Raven *et al.* 1990) is a standardized test that includes 36 items, divided into three subtests of 12 items each. Within each subtest, the items are ordered in increasing difficulty. There was no set time limit to complete the items, and all participants were encouraged to complete all 36 items. Testing took place on a one-to-one basis in a quiet room and was performed according to the manual's instructions. The RCPM was chosen, not only for its simplicity and speed of administration and scoring, but primarily because it has been extensively used for assessment of the fluid-like component of intelligence of typical and clinical populations of children (Cotton *et al.* 2005; Pueyo *et al.* 2008; Van Herwegen *et al.* 2011).

Statistical analysis

In order to compare motor functions and tactile localization abilities, the children were divided into two groups according to MACS and GMFCS levels. Nine children, each who scored at level I and greater than level II in the MACS were identified as the high and low U/E motor function groups, respectively. For the L/E scores, nine children, each who scored at level I and greater than level II in the GMFCS were similarly categorized into — the high and low L/E motor function groups (Table 1), respectively. The difference between GMFCS or MACS level I and II is the presence or absence of limitations in their daily life .

After confirmation of the normal distribution of data by Shapiro–Wilk test, the Welch's *t*-test for unequal variances was used to compare all outcome measures between the two groups. Subsequently, the Spearman's rank correlation test was used to determine the correlation between TLT scores and motor function indexes in terms of U/E and L/E, respectively. Following correlation analysis, a forward stepwise multiple regression analysis was performed to identify the variables that best predicted the motor function measures. The dependent variable was the motor outcome and the independent variables were age, intelligence, and TLT score. We also compared finger TLT and the toe TLT scores.

All statistical analyses were performed using SPSS software for Windows, with the level of significance set at $p < 0.05$.

Results

Comparisons between groups

We measured and analyzed the mean outcome values of both body sides, because the participants' dominant hand was not always clear. For upper extremities, the high motor function group had a significantly higher pegboard test scores than those of the low motor function group (t [10] = 5.18, $p < 0.001$) (Table 2). There were no significant differences in age, RCPM score, touch speed, and error, as well as three and five-finger TLT scores between the groups (Table 2).

In reference to the lower extremities, the high motor function group had significantly higher scores in all motor outcomes, GMFM (t [16] = 3.92, $p < 0.01$), one-leg standing (t [10] = 4.53, $p < 0.01$), and one-leg hopping (t [8] = 4.59, $p < 0.01$), compared to those of the low motor function group. Although no significant differences in the three and five-toe TLTs were observed between the groups, there was a significant difference in L/E TLT between the two groups (t [14] = 4.71, $p < 0.001$) (Table 2). In general, the L/E low motor function group had a tendency for misjudgment during the L/E TLT.

Table 2. Comparisons between the high and low motor function groups

	High motor function group (n = 9) (mean ± SD)	Low motor function group (n = 9) (mean ± SD)	p-value
Upper limbs			
Age (mo)	78.6 ± 23.6	87.3 ± 42.5	p = 0.73
RCPM	22.8 ± 7.2	18.1 ± 5.0	p = 0.13
Purdue Peg board test	8.78 ± 2.43	4.33 ± 0.87	$p < 0.001$
Touch speed time (s)	8.29 ± 2.03	10.34 ± 2.23	p = 0.06
Touch error	0.61 ± 0.24	1.36 ± 1.36	p = 0.14
3-finger TLT (%)	94.4 ± 11.8	95.1 ± 9.4	p = 0.90
5-finger TLT (%)	81.1 ± 14.5	67.2 ± 20.0	p = 0.11
Lower limbs			
Age (mo)	86.4 ± 30.3	76.4 ± 37.5	p = 0.62
RCPM score	21.3 ± 6.4	19.6 ± 6.7	p = 0.58
GMFM	92.6 ± 3.7	51.7 ± 31.1	$p < 0.01$
One-leg standing (s)	8.14 ± 4.44	1.11 ± 1.41	$p < 0.01$
One-leg hopping	9.72 ± 6.24	0.17 ± 0.35	$p < 0.01$
3-toe TLT (%)	81.5 ± 15.2	77.8 ± 21.0	p = 0.67
5-toe TLT (%)	56.1 ± 10.5	56.1 ± 14.5	p = 1.0
L/E TLT (%)	95.8 ± 8.8	74.4 ± 12.0	$p < 0.001$

Table 3. Correlations between upper extremity motor function measures and age, intelligence, and finger TLT

	Month	RCPM	3-finger TLT	5-finger TLT
Purdue Pegboard test	0.10	0.46	0.19	**0.49***
Touch speed test	**−0.56***	**−0.70****	−0.31	**−0.58***
Touch error	−0.06	0.08	0.42	−0.03

Note: Values marked in bold indicate a significant correlation.
*$p < 0.05$; **$p < 0.01$.

Association between motor function and TLT, age, and intelligence

With regards to the U/E, significant correlations were found between the five-finger TLT and the pegboard test (rs = 0.49, $p < 0.05$) as well as the touch speed tests (rs = −0.58, $p < 0.05$). In addition, touch speed test results were negatively correlated with age (rs = −0.56, $p < 0.05$) and intelligence (rs = −0.70, $p < 0.01$), but not with

Table 4. Results of the forward stepwise regression analyses for the upper extremity tests

Variable	B	SE B	β	t	p-value
Purdue Pegboard test ($R=0.56$, $R^2=0.31$, Adjusted $R^2=0.27$)					
Constant	1.52	1.97			
RCPM	0.25	0.09	0.56	2.69	0.02
Touch speed test ($R=0.64$, $R^2=0.41$, Adjusted $R^2=0.38$)					
Constant	14.05	1.47			
RCPM	−0.23	0.07	−0.64	−3.36	0.004

Table 5. Correlations between L/E motor function measures and age, intelligence, and TLT

	Month	RCPM	3-toe TLT	5-toe TLT	L/E TLT
One-leg standing	−0.03	0.14	0.04	−0.18	**0.70****
One-leg hopping	0.15	0.21	0.24	0.04	**0.69****
GMFM	0.02	0.19	0.07	−0.16	**0.80****

Note: Values marked in bold indicate a significant correlation.
*$p < 0.05$; **$p < 0.01$.

Table 6. Results of the forward stepwise regression analyses for the lower extremity tests

Variable	B	SE B	β	t	p-value
One-leg standing ($R=0.57$, $R^2=0.32$, Adjusted $R^2=0.28$)					
Constant	−10.53	5.85			
L/E TLT	17.66	6.45	0.57	2.74	0.02
One-leg hopping ($R=0.58$, $R^2=0.34$, Adjusted $R^2=0.30$)					
Constant	−16.22	7.85			
L/E TLT	24.67	8.58	0.58	2.88	0.01
GMFM ($R=0.89$, $R^2=0.79$, Adjusted $R^2=0.76$)					
Constant	−4.21	24.23			
3-toe TLT	−76.64	20.10	−0.46	−3.81	0.002
L/E TLT	160.10	23.28	0.82	6.88	<0.001

pegboard test results (Table 3). There were no statistically significant correlations between the three-finger TLT and motor function measures, which indicated a ceiling effect. Stepwise regression analysis showed that RCPM was the only independent predictor of the pegboard and touch speed tests ($\beta=0.56$, $p=0.02$ and $\beta=-0.64$, $p=0.004$, respectively; Table 4).

With regards to the L/E, although there were no correlations between either the toe TLT and motor function, the L/E TLT had strong correlations with all motor function measures (GMFM [rs = 0.80, $p < 0.001$], one-leg standing [rs = 0.70, $p < 0.01$], and one-leg hopping [rs = 0.69, $p < 0.01$]; Table 5). We found no correlations between age, intelligence, toe TLT, and the L/E motor outcomes (Table 5). These results were confirmed by performing partial correlation analysis that controlled the effects of age and intelligence. Stepwise regression analysis showed that L/E TLT was the only independent variable statistically associated with all L/E motor outcome measures (Table 6).

Comparison between finger TLT and toe TLT

The correct response rate in the three-finger TLT was significantly higher than that in the three-toe TLT (mean ± SD: three-finger, 94.8 ± 10.3%; three-toe, 79.6 ± 17.9%; t [27] = 3.11, $p < 0.01$). Additionally, the correct response rate in the three-finger TLT was significantly higher than that in the 5-toe TLT (5-finger, 74.2 ± 18.4%; 5-toe, 56.1 ± 12.3%, t [30] = 3.46, $p < 0.01$).

Discussion

In this study, we investigated the response of children with motor deficits to tactile localization on the fingers, toes, and the L/E region, evaluated the latter's association with motor functions.

There were significant differences in the pegboard test of the upper limbs (based on daily motor function as an indicator of manual dexterity) between the two groups divided according to MACS scores. However, there were no significant differences in finger TLT. In contrast, we found that high five-finger TLT scores were associated overall with manual dexterity and touch speed. These results are consistent with recent studies showing an association between finger localization ability and manual dexterity in children with unilateral CP (Auld *et al.* 2012). The authors found that single finger localization was the strongest contributor to unimanual capacity. Our results also suggested that finger TLT is closely correlated with unimanual performance, as shown by the results of the pegboard and touch speed tests, but not with bimanual performance measured using the MACS.

With regards to the lower limbs, the high L/E function group (GMFCS = 1) showed significantly higher scores compared to those of the low L/E function group (GMFCS > 1) for all motor function measures. Similar to the U/E values, although there were no significant differences in the toe TLT between the groups, we found that the high L/E motor function group had significantly higher scores for only L/E TLT compared to those of the low motor function group. The L/E TLT, but not toe TLT, was positively correlated with motor functions. Furthermore, the result of stepwise regression analysis showed that the only significant predictor of L/E motor outcomes was L/E TLT. This finding suggests that partial recognition, (i.e., a part has been touched—in this case, the toes only) is not sufficient for maintaining standing balance and stability. In other words, it is possible that we require the total limb's body image in order to maintain standing balance, as tactile localization on the map of body parts reflects the overall body image (Anema *et al.* 2009; Lefford *et al.* 1974).

Ayres (1979) proposed that body image or schema is critical to motor planning abilities, and that processing tactile as well as proprioceptive information is of critical importance in body image development. The body image is a system of one's own body perceptions, whereas body schema is a system of processes that constantly regulates posture and movement. Image and schema have complementary roles, and both can often be used to monitor and control posture and movements (Gallagher 2001; Gallagher and Cole 1995). Therefore, the correlation between tactile localization and motor performance suggests the possibility that children with motor coordination disorders have developmental delays in terms of their body image.

The observation that finger TLT improved with age was in agreement with previous studies (Lefford *et al.* 1974; Yoshioka *et al.* 2013). Although the reason for this improvement is not clear, it is likely due to the sensory-motor component of daily activities. Interestingly, Gracia-Bafalluy and Noël (2008) demonstrated that finger differentiation training increased finger gnosis. They proposed that finger intervention improved the participants' internal representation of the fingers and hands. Additionally, we found that toe TLT improved with age and intelligence, which was not observed in L/E TLT. As discussed above, L/E TLT was correlated with motor function measures rather than age and intelligence. Toe TLT may also develop in an activity dependent manner.

Our results provide important clinical implications for the rehabilitation of children with developmental disorders. Therapeutic approaches for motor impairment generally use various motor tasks that demand movement. However, our data emphasize the necessity of recognizing one's own body and successfully forming the body image in addition to movement practice for rehabilitation. Body image may be a necessary prerequisite for motor learning. Our results also indicate that motor deficits in children with developmental disorders are likely due to developmental delay in body image. Therefore, rehabilitation therapists should consider not only motor learning, but also the children's perception of their own body or body representation, in order to improve motor function of children with motor deficits. Furthermore, to the best of our knowledge, this is the first study to investigate toe and L/E tactile localization in children with developmental disorders.

However, this study had some major limitations, including the small sample size, the lack of a control group with typical development, and the suitability of the assessments as indicators of body image. Further research could be directed toward developing a more robust clinically useful assessment of body image. The findings in this study should be interpreted with caution because of these limitations. Since there is a paucity of knowledge on toe or lower limb tactile localization abilities, further study is needed to investigate these parameters in typically developing children. Another limitation was the difficulty in concluding the extent to which the development of body image contributes to the improvement of motor performance. This is a subject for further study.

Conclusion

We demonstrated an association between tactile localization ability as an assessment of body image and motor function. We also found that the L/E tactile localization ability was the only independent predictor to be associated with all L/E motor outcome measures. Our data suggest that an undeveloped body image may be associated with motor dysfunction, although this association may be indirect. Rehabilitation approaches focused on somatosensory as well as physical aspects are needed in order to improve motor function in children with developmental disorders, as body image is formed and honed by sensory-motor experiences.

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Conflicts of interest

The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article. The authors alone are responsible for the content and writing of the paper.

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