



Original Article

## Characteristics of the joint position sense in children with developmental dyslexia

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**Abstract.** [Purpose] Developmental dyslexia is a disorder in which reading and writing of characters is difficult. The present study investigated age-dependent joint position sense of the forearm and wrist and whether children with developmental dyslexia have less joint position sense than typically developing children. [Participants and Methods] The participants were comprised of 84 typically developing elementary school students, 12 university students, and 2 children with developmental dyslexia. Joint position sense was evaluated using the reproduction method based on four tasks. The participants were divided into three age groups. The children with developmental dyslexia were compared with the typically developing children in the same age group. [Results] Significant negative correlations were found between the reproduction error of the typically developing children and that of the university students in most tasks. The children with developmental dyslexia showed increased reproduction error relative to the reproduction error of the typically developing children in the same age group in 4 of the 8 tasks. [Conclusion] The accuracy of the joint position sense improved with development. However, the joint position sense of the children with developmental dyslexia was lower than that of the typically developing children in the same age group. The difficulty in writing experienced by children with developmental dyslexia may be related to joint position sensing impairment due to impaired joint position sense.

**Key words:** Developmental dyslexia, Evaluation of joint position sense, Age-dependent joint sense ability

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### INTRODUCTION

Developmental Dyslexia (DD) is a disorder in which the reading and writing of characters is difficult, even though there is no sense disorder (e.g., auditory and visual sense disorders) and the development of intelligence is normal<sup>1)</sup>. The International Dyslexia Association defines dyslexia as a specific learning disability that is neurobiological in origin, characterized by difficulties with accurate and/or fluent word recognition, poor spelling, and decoding abilities, which typically result from a deficit in the phonological component of language that is often unexpected in relation to other cognitive abilities and the provision of effective classroom instruction<sup>2)</sup>. There is no diagnostic label for dyslexia. It is classified as a specific reading disorder in the International Classification of Diseases 10 by World Health Organization diagnostic criteria<sup>3)</sup>. The American Psychiatric Association diagnostic criteria consider specific learning disorders, such as DD, and autism spectrum disorders (ASD) as neurodevelopmental disorders<sup>4)</sup>.

Green et al.<sup>5)</sup> reported that motor development was lower in ASD than in typically developing children in the same age group. In general, the development of sensory integration can be compared to a pyramid of building blocks<sup>6)</sup>. The base of the pyramid comprises the basic sensory functions of the auditory, vestibular, proprioceptive, tactile, and visual senses, and

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building blocks such as maintaining posture, moving the body, forming body images, and acquiring language communication are piled upon the base. However, children with developmental disabilities are described as having an unstable pyramid; elements cannot be stacked well upon the building blocks for each sensory function, which comprise the foundation, and the shape is distorted.

It is known that phonological awareness is greatly involved in the impaired acquisition of character writing in children with DD. Hashimoto et al.<sup>7)</sup> suggested that the difficulty in writing experienced by children with DD is related to impaired kinesthetic memory and ability. Lauren et al.<sup>8)</sup> also suggested that tactile function is one of the causes of upper limb disability in children with Developmental Coordination Disorder. This kinesthetic memory and ability involve motion images based on the position sense of the joint, and belong to proprioception in the somatic sense. The joint position sense, which recognizes the position information of the joint, is equivalent, and is one of the items that construct the base of the pyramid. From this, we are interested in impairing joint position sense affects the severity of reading and writing disorders.

In a previous study, Kagerer et al.<sup>9)</sup> examined the reproducibility of a set joint position of the arm in healthy children from 5 to 14 years of age, and clarified that the accuracy of the joint position sense of the arm improves with development. Pacey et al.<sup>10)</sup> also reported that the joint position sense of the knee joints in children with attention-deficit/hyperactivity disorder was lower than that of typically developing children. However, there are few reports that evaluate the joint position sense of forearm supination and pronation, and wrist joint dorsiflexion and palmar flexion, which are necessary for writing. In addition, the methods of previous studies may be influenced by other somatosensory stimuli by wearing an instrument on the upper extremity.

In the present study, we designed a noncontact method for evaluating the joint position sense of the forearm and wrist. The purpose of this study was to clarify the development of joint position sense of the forearm and wrist according to age in typically developing children and university students, and to evaluate the joint position sense of the forearm and wrist in children with DD relative to that in typically developing children in the same age group.

## PARTICIPANTS AND METHODS

The participants comprised 72 typically developing elementary school students (36 boys and 36 girls) who used an after-school care program, 12 university students (7 males and 5 females; age,  $22.1 \pm 6.4$  years), and 2 children with DD (2 boys, aged 8 and 11 years) who regularly visited a medical institution. Individuals with intellectual disabilities and neurological or orthopedic problems were excluded.

Joint position sense was assessed using a reproduction method based on the report of Goble<sup>11)</sup>. In this method, participants closed their eyes and attempted to reproduce a set target position within 5 seconds. We performed four target joint angles (forearm pronation and supination, and wrist palmar flexion and dorsiflexion). The target joint angles were set to  $30^\circ$  and  $60^\circ$  using a goniometer or inclinometer, and two evaluations were performed for each target joint angle. Measurements were taken using a smartphone (iPhone 7, Apple, USA) fixed to a tripod. Differences between the target and reproduced joint angles were calculated from photographed images using the analysis software, Kinovea ver.0.8.25 (Kinovea, Bordeaux, France). The reproducibility error was defined as the absolute value of the difference between the target joint angle and the reproduced joint angle.

For the forearm pronation and supination tasks, the forearm was placed on the table in the sitting position, holding a 30-cm plastic rod, tilted in accordance with the set angle. For the wrist palmar flexion and dorsiflexion tasks, the forearm was placed on the table in the sitting position, and the joint was moved according to the set angle. Four points were used as landmarks: the styloid process, mid-radius, proximal end of the scaphoid and distal end of the second metacarpal.

Developmental effects in reproducibility error were first evaluated according to school grade. The Shapiro-Wilk test was used to evaluate data normality, which could not be confirmed. Thus, differences in reproducibility error were also evaluated between three age groups: the lower-grade group (41 students: 26 boys; age,  $7.7 \pm 11.0$  years) comprised students from the first to third grades of elementary school, the upper-grade group (31 students: 10 boys; age  $11.0 \pm 9.9$  years) comprised students from the fourth to sixth grade of elementary school, and the university group comprised university students. It should be noted that Japanese elementary schools have a 6 year system, and the age range of the first grade is 6–7 years. The Kruskal-Wallis test, followed by multiple comparisons and adjustment of the significance level using the Dunn-Bonferroni test were used to evaluate differences in reproducibility error between typically development children (first to sixth grade) and university students for each target joint angles. Spearman's rank correlation coefficient was used to examine the relationship the reproducibility error between typically development children and university students for each target joint angles. Furthermore, the reproducibility error values for children with DD were compared to those of typically developing children in the same age group; deviation values were set to 1.5 times the interquartile range (IQR) of each age group. Statistical analyses were performed using SPSS version 21.0 (IBM, Armonk, NY, USA). Statistical significance was recognized at  $p < 0.05$ .

This study was approved by the Ethics Review Board of the International University of Health and Welfare, and informed consent was obtained after providing an explanation of the study to the participants and guardians (approval number 15-T-13).

## RESULTS

The reproduction error in each target joint angle is shown in Table 1. The reproduction error significantly differed between pronation at 30°, pronation at 60°, supination at 60°, and palmar flexion at 60° for all school grades. There was a significant difference between the first-graders and university students in the reproduction error for pronation at 60° and palmar flexion at 60°. In contrast, there were no significant differences between school grades in the reproduction error for supination at 30°, palmar flexion at 30°, dorsiflexion at 30°, and dorsiflexion at 60°. As the school grade increased, the reproduction error tended to decrease. In addition, we found a significant negative correlation between the reproduction error and school grade for each task, with the exception of supination at 30°.

In Table 2, the values of the reproducibility errors for the typically developing children were divided into two groups, lower and upper grades, and were compared to the children with DD of the same grade. The reproduction errors of the with DD in a lower grade were higher than 1.5 times the IQR of typically developing children in the same age group for three conditions: palmar flexion at 60°, dorsiflexion at 30°, and dorsiflexion at 60°, and was higher than the third quartile for

**Table 1.** Analysis of differences in reproduction error according to school grade

Target joint angles	Reproduction error (25–75%ile)							Comparison between grade (p)	Correlation between grade (ρ)
	First grade	Second grade	Third grade	Fourth grade	Fifth grade	Sixth grade	University students		
Pronation 30°	5.5 (3.5–9.1)	4.6 (4.3–9.4)	3.9 (2.3–5.5)	3.2 (2.5–6.1)	4.3 (3.2–5.8)	4.7 (2.8–7.0)	2.3 (1.6–3.7)	0.01	–0.43††
Pronation 60°	9.4 (4.2–16.8)	8.3 (4.6–10.1)	8.0 (3.3–12.7)	4.4 (1.9–7.2)	4.2 (3.5–5.3)	5.1 (3.1–10.0)	2.6* (2.2–3.2)	0.00	–0.47††
Supination 30°	5.4 (4.9–8.0)	7.0 (5.1–8.8)	5.1 (3.1–9.1)	3.4 (2.0–4.7)	5.2 (2.9–7.7)	4.1 (0.9–8.4)	7.0 (3.8–7.9)	0.11	–0.08
Supination 60°	10.2 (6.1–12.4)	7.1 (3.5–9.0)	3.6 (2.3–5.5)	4.3 (2.9–6.3)	2.5 (1.3–5.1)	4.3 (2.4–6.9)	3.0 (1.9–4.5)	0.00	–0.44††
Palmar flexion 30°	8.5 (7.3–9.3)	7.7 (5.2–9.7)	6.1 (2.5–7.1)	5.7 (3.8–9.5)	6.1 (2.1–8.4)	4.8 (1.3–5.3)	4.0 (2.4–7.6)	0.05	–0.28†
Palmar flexion 60°	6.7 (4.1–14.3)	5.8 (3.4–6.6)	5.4 (3.1–6.3)	3.0 (1.3–4.4)	6.0 (2.7–10.7)	2.8 (2.0–6.8)	2.8** (2.1–3.5)	0.00	–0.42††
Dorsiflexion 30°	5.0 (3.9–7.8)	4.9 (2.7–6.4)	3.3 (2.4–5.4)	3.6 (2.0–8.4)	2.9 (2.0–5.8)	3.5 (1.6–5.3)	3.3 (2.5–3.7)	0.26	–0.25†
Dorsiflexion 60°	4.0 (3.2–5.5)	4.4 (2.3–7.9)	4.2 (2.7–6.8)	4.3 (2.6–6.6)	3.4 (2.1–6.4)	5.1 (1.7–6.7)	2.5 (1.5–2.8)	0.06	–0.33††

Significantly different from first grade vs. university student (\*p<0.05, \*\*p<0.01), significantly correlation between each target joint angle (†p<0.05, ††p<0.01).

**Table 2.** Comparison of reproducibility errors between children with DD and typically developing children in the same age group for each target joint angles

Target joint angles	Children with DD		Typically developing children			
	Lower grade n=1	Upper grade n=1	Lower grade n=41	75%ile (75%ile+IQR *1.5)	Upper grade n=31	75%ile (75%ile +IQR *1.5)
Pronation 30°	11.2†	1.2	5.25	6.5 (11.5)	4.61	6.0 (10.8)
Pronation 60°	7.1	4.1	10.05	12.8 (25.8)	5.09	6.8 (12.4)
Supination 30°	1.8	8.3†	6.55	8.8 (14.6)	4.70	6.4 (12.4)
Supination 60°	6.7	7.7†	7.16	10.2 (20.4)	4.37	5.9 (11.2)
Palmar flexion 30°	7.4	9.1†	7.10	9.3 (16.7)	5.54	8.1 (16.1)
Palmar flexion 60°	11.9††	7.6†	6.93	7.0 (11.6)	4.37	6.1 (12.0)
Dorsiflexion 30°	21.2††	1.4	4.84	6.4 (11.9)	4.44	5.9 (11.6)
Dorsiflexion 60°	12.9††	5.5	5.02	6.7 (12.4)	4.63	6.3 (12.2)

DD: Developmental dyslexia; 75%ile: The third quartile percentile; IQR: Interquartile range; †75%ile<Reproduction error, ††75%ile+IQR\*1.5 <Reproduction error.

four conditions. The reproduction errors of the child with DD in an upper grade were higher than third quartile of typically developing children in the same age group for four conditions: supination at 30°, supination at 60°, and palmar flexion at 30° and 60°. Thus, children with DD in both the lower and upper grades showed increased reproduction errors in 4 of the 8 tasks compared to that in typically developing children in the same age group.

## DISCUSSION

Joint position sense is often evaluated in large joints, such as the knee joint for total knee arthroplasty<sup>12)</sup>. In general, joint position sense changes in development, increasing in accuracy. For example, Kagerer et al.<sup>9)</sup> clarified that the accuracy of the joint position sense of the arm improved with development. Furthermore, Holst-Wolf et al.<sup>13)</sup> evaluated the joint position sense of the forearm and reported a change in accuracy with development. However, these studies used evaluation methods that are carried out by placing a measurement instrument in contact with the body; thus, the measurements are affected by tactile and pressure-sense information. In the present study, we focused on the method reported by Tsushima et al.<sup>14)</sup> to obtain accurate joint angle from static images and tried a non-contact method to evaluate the joint position sense of the forearm and wrist. The advantage of this method is that other somatosensory inputs can be excluded during the evaluation. Using a non-contact method of evaluation, we showed that the reproducibility error in joint position sense decreased with increasing age. In other words, the accuracy of the joint position sense improved with development. Accordingly, the reproducibility error decreased as the school grade (as well as age group) increased for all target joint angles, with the exception of supination at 30°. The reason for this result is that the joint position sense is required to a greater extent and the frequency of writing increases with increasing school grade. Rueckriegel et al.<sup>15)</sup> reported that the motor skills of the fingers improved with age. Furthermore, Erhardt et al.<sup>16)</sup> reported that with development, the dominant sensations change from touch, pain, and thermal nociception sensations to sensations related to position and movement. These results are consistent with the improvement in the joint position sense with development clarified in the present study.

The reproducibility error of the joint position sense was increased in children with DD relative to that in children of the same age. This result suggests that children with DD have reduced joint position sense. The child with DD in the lower grade had reproducibility errors that deviated from normal (values greater than 1.5 times the IQR or in the third quartile percentile of typically developing children in the same age group) in 4 out of 8 tasks: pronation at 30°, palmar flexion at 60°, dorsiflexion at 30°, and dorsiflexion at 60°. In addition, the child with DD in the upper grade had reproducibility errors that deviated from normal (values greater than the third quartile percentile of typically developing children in the same age group) in 4 out of 8 tasks: supination at 30°, supination at 60°, palmar flexion at 30°, and palmar flexion at 60°. Thus, in the present study, increased reproducibility errors were observed in 4 out of 8 tasks in children with DD compared to that in healthy children in the same age group for both lower and upper grades. These results suggest that impaired joint position sense is involved in children with DD who have difficulty acquiring letters. There are two Kanji learning methods, learning writing conditions and writing like finger movements in Japanese. In both cases, the muscles of the hand are effectively used to perform muscular and kinetic writing<sup>17)</sup>. It has also been reported that the kanji learning process is based on a visual analysis of the components of Kanji, and realizes writing while stimulating the motor memory necessary for writing<sup>18)</sup>. Writing like finger movements<sup>17, 19)</sup> and finger-tracing<sup>20, 21)</sup> are thought to be effective in the formation of motor memory. Information from the joint position sense necessary for the writing procedures of the forearm and wrist seems to be significantly related to the formation of motion memory. The decrease in joint position sense in children with DD revealed in the present study is considered to be one of the causes of the difficulty in acquiring letters.

The method for evaluating the joint position sense proposed in the present study utilized easily available materials, without special inspection equipment. As a result, it was possible to construct an evaluation system that could be purchased at a nearby store, with little cost. This is a merit of this method for clinical applications. A disadvantage of this method is that it takes more than 30 minutes to evaluate 8 exercise tasks performed 2 times each.

The present study has the following limitations. The study had a small number of participants, with a mismatch in the number of participants between groups; unequal education by regional differences; and age differences within the school grade (i.e. early and late births within the school grade). In the future, the relationship between the development of the joint position sense and the disease state of children with DD should be further clarified in larger samples with a wider age range (such as junior high school and senior high school students). In addition, the relationship between the dominant and non-dominant hand should be clarified by evaluating the joint position sense of the non-dominant hand. Furthermore, for future practical applications of our proposed evaluation method, it is necessary to collect more sample data and establish the inter-rater reliability. For a more accurate measurement, it is necessary to decrease the number of exercise tasks and increase the number of procedures.

In this study, the accuracy in joint position sense improved with development. However, the joint position sense of children with DD was lower than that of typically developing children in the same age group. These results suggest that a decrease in joint position sense in the two children with DD in this study may be one of the causes of their difficulty in acquiring letters.

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### *Conflict of interest*

The authors declare no conflicts of interest associated with this manuscript.

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