ORIGINAL REPORT



JOINT POSITION SENSE OF LOWER EXTREMITIES IS IMPAIRED AND CORRELATED WITH BALANCE FUNCTION IN CHILDREN WITH DEVELOPMENTAL COORDINATION DISORDER

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Objective: To determine the joint position sense of the lower extremities and its relationship with motor function in children with developmental coordination disorder and typically developing children.

Methods: A total of 56 participants were recruited; 28 children with developmental coordination disorder (mean age 10.86 years (standard deviation (SD) 1.07); 13 females, 15 males) and 28 typically developing children (mean age 10.96 years (SD 1.18); 12 females, 16 males). Knee and ankle joint position sense were assessed using a Biodex isokinetic dynamometer. Joint position acuity was measured by position error (PE) and position error variability (PEV). Motor function was examined using the 2nd edition of Movement Assessment Battery for children (MABC-2) and quantified via sub-scores from 3 MABC-2 domains.

Results: Both PE and PEV at knee and ankle joints were significantly greater in children with developmental coordination disorder compared with typically developing children. In addition, both PE and PEV were significantly greater at the ankle joints than the knee joints in children with developmental coordination disorder. For children with developmental coordination disorder only, joint position acuity in the lower extremities significantly and negatively correlated with MABC-2 balance sub-score.

Conclusion: This study verifies that lower limb proprioception is impaired in children with developmental coordination disorder. Also, children with developmental coordination disorder displayed greater proprioceptive deficits at the ankle compared with the knee joint. Children with developmental coordination disorder who had poorer joint position acuity, i.e. greater PE and PEV, in the lower extremities tended to perform less well in balance function.

Key words: joint position sense; balance function; knee; ankle; developmental coordination disorder.

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Developmental coordination disorder (DCD) is a common neurodevelopmental motor condition with a prevalence of 5–6% in children ages 5–11 years

LAY ABSTRACT

This is the first study to investigate joint position sense (the awareness of the postion of a joint in space) of the lower limbs and its relationship with motor function in children with developmental coordination disorder and typically developing children, while addressing limitations in previous studies. The results show that children with developmental coordination disorder have: (i) impaired joint position sense at both knee and ankle joints; (ii) poorer joint position sense at ankle than knee joints; and (iii) decreased balance function with increasing impairment in joint position sense in their lower extremities. These findings suggest that lower limb proprioception is impaired as well as joint position acuity of lower extremities plays a role in balance function in children with developmental coordination disorder.

worldwide (1). The motor coordination deficits of DCD crucially interfere with activities of daily living (e.g. buttoning, tying shoelaces, using utensils), as well as poor participation and performance in school (e.g. painting, handwriting, sports) (1). Children with DCD show coordination difficulties across a variety of motor domains, such as manual dexterity, aiming and catching, and balance (2). To date, the aetiology and pathogenesis of DCD are unknown. Proprioceptive information does not only affect the detection and correction of movement errors, regulation of coordinated movement, motor learning, and balance control, but also impacts individual function and independence in daily life (3, 4). Therefore, the current study was particularly concerned with testing proprioceptive sense and exploring its relationship with motor function in children with DCD.

Recently, studies have begun to adopt a joint position matching paradigm to evaluate proprioceptive characteristics in children with DCD. One study using an "active" matching test reported that the joint position sense of both elbows and wrists is impaired in children with DCD compared with typically developing (TD) children (5). Also, the study unexpectedly demonstrated that elbow/wrist joint position sense was not correlated with motor function in either the DCD or the TD groups. Similarly, another study employing an identical test protocol found significantly poorer proprioceptive acuity of the wrist joints in children with DCD compared with TD children, and no correlation

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between wrist joint position sense and motor function between the DCD and TD groups (6).

In these studies, upper extremity joints were moved passively from the neutral joint position to a target position by an experimenter, at an approximate velocity of 20–25°/s, and held there for 3 s before returning to the neutral position. Children were then required to actively reproduce the target position with no visual or auditory feedback. However, several factors should be considered in this "active" joint position matching test. Firstly, limb velocity was passively and manually controlled by an experimenter and actively moved by the children themselves. Joint angular velocity can impact proprioceptive sense, with higher joint velocities showing decreased detection threshold (increased sensitivity) to proprioception, and vice versa (7, 8), varied limb movement velocity can confound the perception of joint position sense. Secondly, an "active" matching test using volitional control of limb movement showed that, during active movement, an efferent copy of the motor command is yielded and transmitted to the central nervous system (CNS) to anticipate the consequences of the movement itself (9). In this case, the active joint position matching test is not a "pure" test of proprioceptive sense, and matching test performance is influenced by the integration of both internal prediction information and external sensory signals (10). Furthermore, a core feature of DCD is poor motor control, as shown in altered movements and postures leading to abnormal motor function (e.g. 11, 12). This means that the active joint position matching test may be suboptimal for estimating proprioception. Lastly, to the best of our knowledge, no research has yet investigated proprioception in the lower extremities using a joint position matching paradigm in children with DCD. Furthermore, little is known about the specific association between joint proprioceptive acuity in the lower extremities and motor function in children with and without DCD.

Hence, the objectives of this study were to determine the joint position sense of lower extremity joints, as well as correlations between lower extremity joint proprioceptive acuity and motor function performance in children with DCD and TD children. The study controlled for suspected confounding factors, such as inconsistent joint angular velocity and active control of limb movement.

METHODS

Written informed consent was obtained from all participants and their parents, and ethics approval was provided by the Institutional Review Board at Antai-Tian-Sheng Memorial Hospital in Pingtung County, Taiwan (IRB No. 19-020-A). The study methods complied with the principles of the Declaration of Helsinki for human subject research.

Participants

A total of 56 participants, age range 9–12 years, were recruited from local schools; 28 children with DCD (7 in each age group; mean age 10.86 years (standard deviation (SD) 1.07); 13 females, 15 males) and 28 TD children (7 children in each age group; mean age 10.96 years (SD 1.18); 12 females, 16 males). Motor function was assessed with the 2nd edition of Movement Assessment Battery for Children (MABC-2), which has 4 age bands, each with 8 test items nested in 3 motor domains (manual dexterity, aiming and catching, and balance) (13). In accordance with the MABC-2 manual, the raw score of each item was converted to a standard score based on normative data, and the MABC-2 total score was the sum of the standard sub-scores of the 3 domains. For children with DCD, motor coordination and balance function were significantly below that expected for their age, as confirmed by a < 5th percentile total score and a < 5th percentile balance subscore from MABC-2. In addition, in children with DCD motor coordination difficulties substantially interfered with activities of daily living and school activities, as confirmed by a>95th percentile score on the checklist of MABC-2. TD children had a>15th percentile total score and a>15th percentile balance subscore on the MABC-2 test, as well as a < 85th percentile score for the MABC-2 checklist. Both the DCD and TD groups were not comorbid for ADHD (Attention-deficit/hyperactivity disorder), as confirmed by a < 70 score on Conners' Teacher Rating Scale (CTRS) (14). All participants showed normal intelligence, as confirmed by a>80 score on the 2nd edition of the Kaufmann Brief Intelligence Test (KBIT-2) (15). A history of severe lower extremity injuries, e.g. muscle tear, ligament rupture, or fracture, or neurological disorders, e.g. epilepsy, cerebral palsy, and

Table I. Basic characteristics of the children with developmental coordination disorder (DCD) and typically developing (TD) children

Measure	Children with DCD (n=28; 13 girls, 15 boys) (7 children in each age group)	TD children $(n=28; 12 \text{ girls}, 16 \text{ boys})$ $(7 \text{ children in each age group})$	t	<i>p</i> -value
Age, years, range	9-12	9–12	-	-
Age, years, mean (SD)	10.86 (1.07)	10.96 (1.18)	0.34	0.74
Height, cm, mean (SD)	ht, cm, mean (SD) 142.25 (5.45)		-0.54	0.59
Weight, kg, mean (SD)	41.93 (4.20)	40.14 (4.70)	-1.50	0.14
KBIT-2, mean (SD)	mean (SD) 102.79 (9.23)		-0.31	0.76
CTRS, mean (SD) 48.00 (5.70)		48.96 (5.82)	0.63	0.53
MABC-2 total score, mean (SD)	ABC-2 total score, mean (SD) 50.54 (4.93)		17.36	0.00
Manual dexterity sub-score, mean (SD)	19.70 (1.75)	24.29 (2.61)	7.73	0.00
Aiming and catching sub-score, mean (SD)	17.11 (3.82)	18.89 (2.73)	2.01	0.05
Balance sub-score, mean (SD)	13.73 (3.17)	28.36 (3.05)	17.61	0.00

KBIT-2: Kaufmann Brief Intelligence Test 2nd edition; CTRS: Conners' Teacher Rating Scale; MABC-2: Movement Assessment Battery for Children 2nd edition.

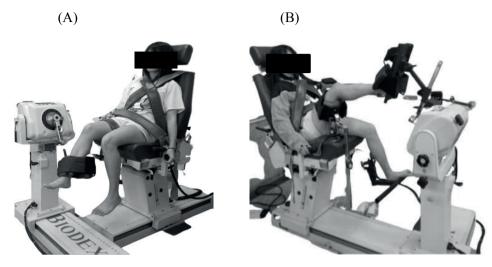


Fig. 1. Experimental setup for joint position matching for: (A) knee joint and (B) ankle joint. Permission has been given to publish the photo from the patients

Tourette's syndrome, which may affect children's motor function and development, were excluded based on parents' reports. All participants were right-foot dominant, as determined by the preferred leg used to kick a ball (16). Table I shows the basic data for the children with DCD and TD children.

Procedures

The study was conducted at the Sport Biomechanical Laboratory, National Kaohsiung Normal University, Kaohsiung City, Taiwan. The laboratory was kept quiet and free from distraction throughout the experimental procedure. Details of the experimental procedures were explained to participants by a researcher before commencing the study. Participants were seated on a Biodex isokinetic dynamometer (Biodex System 3, Biodex Medical Systems Inc., Shirley, NY, USA), leaning their back against a rigid backrest (Fig. 1). Subjects were stabilized with foam rubber straps across the trunk and hip joints. Participants were asked to wear sport shorts with no shoes during the execution of the "passive" ipsilateral joint position matching test. To prevent mental fatigue resulting from the prolonged test time, only the dominant leg was assessed. Children were blindfolded to eliminate external visual cues. They also listened to pink noise through headphones, to mask external auditory cues.

To measure knee joint position sense, the Biodex dynamometer was adjusted so that participants were seated in the chair (backrest inclined at 85° from horizontal), with the hip flexed at 90° and the popliteal fossa situated approximately 5 cm from the edge of the seat. The participant's thigh was strapped, and the lower leg was secured to the lever arm immediately above the lateral malleolus. The rotational axis of the dynamometer lever arm was aligned with the knee joint axis of flexion and extension (lateral femoral epicondyle) with a knee flexion at 90° (Fig. 1A). To measure ankle joint position sense, participants were seated in the chair (backrest inclined at 70° from horizontal), with the hip flexed at 90° and knee flexed at 20°, while the foot was fixed to the footplate using Velcro and elastic straps. The rotational axis of the dynamometer lever arm was positioned to be coaxial with the ankle joint axis of plantarflexion and dorsiflexion (fibular malleolus), with ankle dorsiflexion at 0° (Fig. 1B).

To discourage participants from using timing cues, the starting position and target angle were not constant among test trials, based on previous work (17-20). To begin, the participants' limb was passively moved from a starting position (knee flexed 80°, 90°, or 100° for the knee matching test; ankle dorsiflexed at 0°, 5°, or plantar flexed at 5° for the ankle matching test) to a target angle (30° or 60° flexion of knee joint and 10° or 15° dorsiflexion of ankle joint in an open kinetic chain by the isokinetic dynamometer). The joint angular velocity was set to a constant 1°/s. The presentation order of the combined starting position and target angle varied across participants following a Latin square design. The limb was then maintained at the target angle for 5 s to allow children to memorize the position. Afterwards, the limb was returned to the starting position. The dynamometer then again passively moved the leg from the starting position toward the target angle and participants were instructed to replicate the target angle as closely as possible. Participants had to indicate when the "perceived" target angle had been reached, by pressing a button to stop the motion of the dynamometer. Participants could attempt to fine-tune the perceived angle until they were satisfied with their adjustments and matches. Subsequently, participants' lower limb was passively returned to the starting position for the next trial. No verbal feedback was provided by the experimenter.

There were 20 trials in total, 10 for the knee joint position matching test (5 for each target angle) and 10 for the ankle joint position matching test (5 for each target angle). Trials were blocked and counter-balanced, such that half the children with DCD and TD children first performed the knee joint matching test, followed by the ankle joint matching test. The other half of the subjects performed the tests in the reverse order. There was a 10-s inter-trial interval and a 10-min resting interval between blocks. Prior to formal measurements, 2 practice trials confirmed that all participants were familiar with the testing procedures. If any participants seemed unsure, additional instruction, demonstration, and practice were provided. Participants took approximately 30–40 min to complete the entire experimental procedures. Fig. 2 shows the experimental procedures.

Statistical analysis

All measurements were summarized as means with standard deviation (SD). Dependent variables were basic data and

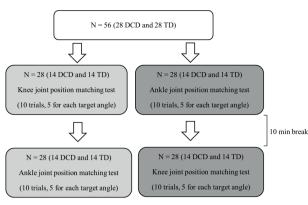


Fig. 2. Experimental procedures. DCD: developmental coordination disorder; TD typically developing children.

proprioceptive acuity data, including position error (PE) and position error variability (PEV). The PE was calculated by taking the absolute value of the difference between the perceived angle and target angle in degrees (formula 1). The PEV was calculated as SD of the PE across trials (formula 2). Normality and homogeneity of variance were checked using Kolmogorov-Smirnov and Levene's tests. Basic data were compared using an independent t-test. Proprioceptive acuity data (PE and PEV) were analysed using a Group (2 levels: children with DCD and TD children) × Joint (2 levels: knee and ankle) mixed analysis of variance (ANOVA) model. Significant interactions were further investigated using the Bonferroni post-hoc analysis to demonstrate the simple main effects of either Group or Joint. Partial eta-squared (n, 2) was calculated for ANOVA effects to indicate effect size. Pearson product-moment correlations were conducted between proprioceptive acuity data (PE and PEV) and sub-score in 3 motor domains of the MABC-2 (manual dexterity, aiming and catching, and balance) for children with DCD and TD children, separately. Significance was accepted for p-values < 0.05. Statistical analysis was performed with SPSS version 18.0 software package (SPSS Inc., Chicago, IL, USA).

PE=|P-T|

Formula 1. Calculation of PE measures, where P=degree of the perceived joint angle; T=degree of the target joint angle.

 $PEV = \sqrt{\frac{\sum_{i=1}^{N} (P_{i}-P)}{\sum_{i=1}^{N} (P_{i}-P)}}$

Formula 2. Calculation of PEV measures, where N=total number of trials; Pi=degree of the perceived joint angle on trial i; P,=mean degree of the perceived joint angle.

RESULTS

Basic data

No significant differences were found between groups for age, height, weight, or scores on CTRS and KBIT-2 (p>0.05). However, the DCD group were significantly lower than the TDC group for the manual dexterity sub-score (t(54)=10.82, p<0.05), aiming and catching sub-score (t(54)=15.13, p<0.05), balance sub-score (t(54)=12.29, p<0.05), and total score for MABC-2 (t(54)=21.68, p<0.05).

Proprioceptive acuity data

Fig. 3 demonstrates the data regarding PE for the 2 groups of participants at both knee and ankle joints. In terms of PE, the ANOVA identified an interaction effect between Group and Joint (F (1, 54) = 18.23, p < 0.05, np2 = 0.25). Bonferroni post-hoc analysis revealed a significant simple main effect of Group for the knee joint (F (1, 54)=-2.85, p=0.01) and ankle joint (F (1, 54)=-10.37, p<0.01), indicating that DCD children showed larger position error compared with TD children for both knee (DCD=6.21±3.45°, $TD = 4.27 \pm 1.06^{\circ}$) and ankle joints (DCD = 8.77 \pm 2.19°, TD=3.73±1.36°). Furthermore, Bonferroni post-hoc analysis revealed a significant simple main effect of joint in the DCD group (F (1, 54)=-4.01, p<0.01), but not in the TD group (F (1, 54) = 1.57, p = 0.13), indicating a larger position error in the ankle than in the knee joint for the DCD group only (knee = $6.21\pm3.45^{\circ}$, ankle=8.77±2.19°). Detailed PE data are provided in Appendix I.

Fig. 4 shows data regarding PEV for 2 groups of participants for both knee and ankle joints. For the PEV, ANOVA identified an interaction effect between Group and Joint ($F(1, 54) = 16.72, p < 0.05, \eta_p^2 = 0.24$). Bonferroni *post-hoc* analysis revealed a significant simple main effect of Group at the knee joint (F (1, 54)=-2.73, p=0.10) and at the ankle joint (F (1, 54)=-9.90, p<0.01), indicating that DCD children showed larger position error variability compared with TD children for both knee (DCD=2.92±1.66°, TD=2.02±0.50°) and ankle joints (DCD=4.31±1.01°, TD=1.91±0.78°). In addition, Bonferroni *post-hoc* analysis revealed a significant simple main effect of joint in the DCD group (F (1, 54)=-4.53, p<0.01).

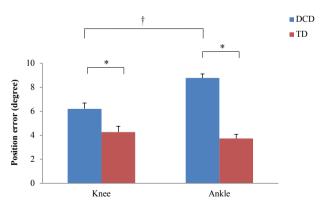


Fig. 3. Position error of knee and ankle joints in children with developmental coordination disorder (DCD) and typically developing (TD) children. Asterisks denote significant differences in position error between the DCD and TD groups. Daggers denote significant differences in position error between knee and ankle joints within the DCD group. Error bars show standard deviation.





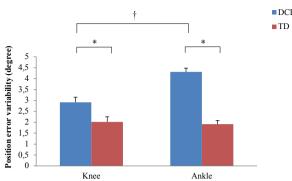


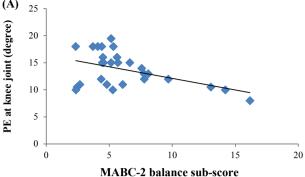
Fig. 4. Position error variability of knee and ankle joints in children with developmental coordination disorder (DCD) and typically developing (TD) children. *Significant differences in position error variability between the DCD and TD groups. †Significant differences in position error variability between knee and ankle joints within the DCD group. Error bars show standard deviation.

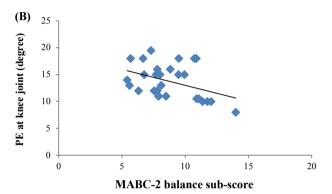
but not in the TD group (F (1, 54)=0.61, p=0.55), indicating larger position error variability in the ankle than in the knee joint for the DCD group only (knee= $2.92\pm1.66^{\circ}$, ankle= $4.31\pm1.01^{\circ}$). Detailed PEV data are shown in Appendix II.

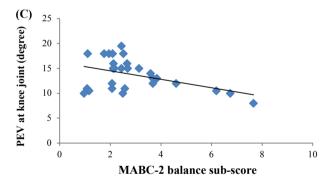
Fig. 5 demonstrates correlations between proprioceptive acuity data (PE and PEV) and MABC-2 balance sub-score for both the ankle and knee joints in DCD children. Within the DCD group, there were significantly negative correlations between PE and MABC-2 balance sub-scores for the knee joint (r=-0.47, p=0.01, Fig. 5A) and ankle joint (r=-0.41, p=0.41)p=0.03, Fig. 5B), as well as significant negative correlations between PEV and MABC-2 balance sub-scores for the knee joint (r=-0.44, p=0.02, Fig. 5C) and ankle joint (r=-0.42, p=0.03, Fig. 5D). However, the proprioceptive acuity data did not significantly correlate with either manual dexterity sub-score (p>0.05) or aiming and catching sub-score (p > 0.05) in children with DCD. Furthermore, within the TD group, no significant correlations were detected (p > 0.05). Detailed correlation data are shown in Appendix III.

DISCUSSION

This study investigated the characteristics of joint position sense in the lower extremities for children with and without DCD. The major findings were that children with DCD have: (i) inferior proprioceptive acuity, measured in terms of position error and position error variability, in the knee and ankle joints; (ii) poorer proprioceptive acuity in the ankle joint compared with the knee joint; and (iii) negative correlations between the impaired joint position sense in their lower extremities and balance function. The results are discussed below.







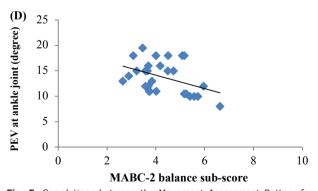


Fig. 5. Correlations between the Movement Assessment Battery for children (MABC-2) balance sub-score and (A) the position error of knee joints, (B) position error of ankle joints, (C) position error variability of knee joints, and (D) position error variability of ankle joints. PE: position error; PEV: position error variability.

Joint position sense acuity is reduced in both knee and ankle joints in developmental coordination disorder

Firstly, the present study demonstrated, for the first time, significantly reduced knee and ankle proprioceptive acuity in children with DCD relative to TD children. Overall, the current study extended the findings of previous studies by demonstrating that proprioceptive deficits in children with DCD is not only localized to upper extremity joints, e.g. elbow and wrist, but also influences lower extremity joints, e.g. knee and ankle. Previous studies observed that the PEV (not PE) of the DCD group was larger compared with the cohort group (5, 6). In line with previous studies, the present study showed that PEV was larger in children with DCD than in TD children, demonstrating a variable perceptual judgment about lower extremity joint position in children with DCD. However, contrary to previous studies, the present study showed that PE was larger in children with DCD compared with TD children, indicating that children with DCD have a reduced ability to identify joint position through proprioception. These variable-dependent differences in proprioceptive performance can be reasonably explained by the different approaches used to determine joint position sense among studies. Previous studies (5, 6) employed an active joint position matching test, which requires voluntary control over skeletal muscle and did not strictly control active and passive limb movement velocity. In contrast, the present study purposely minimized confounding effects from participants' active movement control and altered movement velocity by implementing a passive joint position matching test and applying a constant-velocity mode from the Biodex dynamometer. Based on current and previous studies, for children with DCD, an active joint position matching test with varied movement velocity may not be sensitive enough to detect any proprioceptive deficits, but a passive joint position matching test with constant joint angular velocity is sufficiently sensitive to identify proprioceptive deficits for children with DCD. One may argue that active movements are closely related to activities of daily living, but passive movements are less functionally connected. The question then arises: why does performance on the passive joint position sense test matter? In fact, studies have indicated that a proprioceptive test demanding volitional control of bodily movement may not appropriately reflect the sense of joint position (21). Moreover, anaesthetic block of the joint does not affect active proprioceptive acuity, but merely reduces performance in the passive joint position test, demonstrating that passive joint matching test is more appropriate to examine the

function of proprioceptive receptors in the joint (22). Asthon-Miller et al. (23) and Deshpande et al. (24) also suggested using a test not associated with motor control for measuring changes in proprioceptive acuity. Here, the present study uses a passive matching test to better control for confounding variables when assessing joint position sense, i.e. inconsistent joint angular velocity and active control of limb movement, thus more clearly demonstrating the characteristics of lower extremity proprioceptive deficits in children with DCD.

At present, the mechanism underpinning proprioceptive abnormalities in children with DCD remains poorly understood. Afferent signals coming from both muscle spindles and skin receptors play crucial roles in the sensation of limb movement and position (25). Notably, there is a lack of empirical research concerning the fusimotor sensitivity of muscle spindles in children with DCD. However, a number of studies have shown that children with DCD tend to have hypotonia (26, 27). With low muscle tone, the length of skeletal muscle fibre is longer than normal when the muscle is at rest and/or during a sustained voluntary contraction. Here, the internal force generated by muscle contraction and position or movement of body limbs may not be sufficient to trigger muscle spindle activation. It could, therefore, result in a low firing rate of proprioceptive afferents, which can further lead to joint position sense deficits in children with DCD. Also, joint rotation can result in strains of adjacent skin, i.e. stretched on one side of the joint and slackened on the other side, and stimulate tactile mechanoreceptors in the skin, thus potentially contributing to limb position sensation. Studies have indicated that children with DCD show an elevated threshold for tactile-related sense, e.g. light touch (28) and haptic discrimination (29). A higher tactile-related threshold could, at least in part, account for reduced proprioceptive performance at the knee and ankle joints for children with DCD. Furthermore, similar generalized proprioceptive impairments influencing both knee (30) and ankle joints (31) are seen in patients with Parkinson's disease, which affects the cortico-basal ganglia-thalamo-cortical circuitry. Although there is no direct evidence for anatomical and physiological changes in the sensory area of the brain responsible for proprioception, recent imaging studies reveal that functional connectivity between the motor cortex and primary/secondary somatosensory cortex (postcentral gyrus, parietal operculum, and supramarginal gyri), as well as between the motor cortex and basal ganglia regions (caudate, putamen, and globus pallidus) are altered in children with DCD (32, 33). Future research should systematically investigate the possible causes of impaired proprioception in children with DCD in the peripheral nervous system,

e.g. muscle spindle sensitivity, total number of joint mechanoreceptors, intrafusal, and chain fibres, as well as the central nervous system, e.g. neural activity in the premotor cortex, cerebellum, and the proprioceptive regions of the basal ganglia and grey matter volume in the precentral gyri, postcentral gyri, insula, and angular gyri.

Joint position sense in the ankle is affected more than the knee in developmental coordination disorder only

Secondly, the present study demonstrated differences between joints, with larger PE and PEV in the ankle joint compared with the knee joint in children with DCD. These results corroborate previous studies reporting greater proprioceptive impairment in distal joints than proximal joints in patients with stroke and intracranial disorders (34, 35). The theoretical basis for this proximal-distal disparity may involve altered functional connectivity in regional brain networks. Proprioceptive information from the proximal joint is transmitted to and mediated bilaterally by both the ipsilateral and contralateral hemispheres, while afferents from the distal joint are projected to and processed in the contralateral hemisphere only (36). Thus, the significant difference found in proprioceptive acuity between joints may explain possible lesions in certain afferent pathways and/or atypical activations in the proprioceptive-processing neural area in children with DCD.

Proprioceptive acuity in lower extremity joints negatively correlates with balance function in developmental coordination disorder only

Thirdly, the present study found significant negative correlations between proprioceptive acuity in the lower extremity joints (knee and ankle) and balance function in children with DCD. The ankle joint is crucial in maintaining stability, especially for modulating body sway in the anteroposterior direction (37). Furthermore, the knee joint provides stability while standing upright or on an unstable surface, e.g. foam pads (38). Not surprisingly, the present findings demonstrate that children with DCD who display more impaired joint position sense tend to have worse balance performance. This implies that the proprioceptive status in the lower extremities can predict balance function in children with DCD. However, previous studies did not show correlations between proprioceptive acuity in upper extremity joints (elbow and wrist) and manual dexterity sub-score (5, 6). The question then becomes, why is proprioception in lower extremities significantly associated with gross balance function, but proprioception in upper extremities not related to fine motor skills? A reasonable explanation may be that fine motor skills draw on a wider range of abilities, e.g. visual control, visual perception, and eye hand coordination, so joint position sense in upper extremities contributes less to fine motor performance. Furthermore, the knee joint matching test resembled a sitting position with the angular range required for sit-to-stand. Studies have documented the knee joint largely contributes to lifting body weight during sit-to-stand (39). Without effective knee joint control, i.e. desired trajectory of joint angle, it is difficult to smoothly transfer centre of mass upward from a stable seated position to a relatively less stable standing position, while simultaneously maintaining body equilibrium. Proprioceptive acuity of the knee joint may be related to functional sit-to-stand movement. However, data on sit-to-stand performance were unavailable in this study. Further studies could investigate the relationship between sit-to-stand performance and position sense acuity of the knee joint in children with DCD.

Due to strict inclusion criteria for children with DCD, balance sub-score data were not normally distributed in this study. Considering the inclusion criteria, as well as the high heterogeneity of DCD (2), the results of the current study cannot be generalized to all children with DCD. Also, the balance domain of the MABC-2 is a relatively narrow distribution for the standard score for some tests. Take "hopping on the mat" for example. For the 10–12-year age group, 5 consecutive hops with the preferred leg gives a standard score of 11, while 0-4 hops yields an identical standard score of 1. However, participant ages were spread across 2 age bands of the MABC-2, i.e. age band 2 (7–10 years), and age band 3 (11–16 years). Given the number of participants in each group and that test items differed between age bands, it was not appropriate to compute correlation coefficients for the DCD and TD groups using the raw score for each test item. To further this research line, prospective studies should consider all these aspects, i.e. recruiting various subtypes of DCD, focusing on a single age band for children, and calculating correlation between proprioceptive acuity and raw score on test items for MABC-2.

Limitations and future directions

A major limitation is the possible reliance on working memory. The contralateral joint position test involves matching a reference angle with the opposite limb, which requires no memory demands, whereas the ipsilateral joint position test requires remembering a previous joint angle. The ipsilateral joint position matching test inevitably involves working memory. A previous study suggested the effect of working memory

on ipsilateral joint position matching performance is minimal, as no differences were detected between ipsilateral and contralateral joint position tests (5). Still, from the ipsilateral joint position matching test, we cannot fully exclude working memory as a potential influence in the current study. More studies should investigate proprioceptive acuity in lower extremities using contralateral joint position matching tests in children with DCD. Moreover, no participants were inattentive or distracted during experiments. However, our study did not measure attention bias between groups before and during the proprioceptive test, and the whole experimental procedure lasted 30–40 min, including set-up, instruction, demonstration, practice, and formal test trials. Therefore, whether or not the results were confounded by between-group differences in attention is speculative. Further investigations comparing how attention impacts proprioceptive acuity between children with and without DCD are recommended. Lastly, a variety of interventions, e.g. vibration, manual therapy, taping and bracing, and exercise, have been used to enhance proprioception (23, 40). However, no such interventions have improved proprioception in children with DCD. Furthermore, enhancing joint position acuity does not automatically adapt or facilitate balance performance. Future research should examine which intervention is more effective and efficient in addressing proprioceptive deficits in lower extremities, as well as whether changes in joint position sense promote motor function and skill in lower extremities in children with DCD.

Conclusion

This study verified that impaired joint position sense of knee and ankle joints occurs in children with DCD. In addition, the position error and position error variability in ankle joints is much larger than in knee joints for the DCD group. Lastly, the severity of proprioceptive deficits in knee and ankle joints is negatively associated with balance performance in children with DCD.

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REFERENCES

- American Psychiatric Association. Diagnostic and statistical manual of mental disorders. 5th edn. Washington, DC: American Psychiatric Publishing; 2013.
- 2. Hoare D. Subtypes of developmental coordination disorder. Adapt Phys Act Q 1994; 11: 158–169.
- Carey LM, Oke, LE, Matyas TA. Impaired touch discrimination after stroke: a quantitative test. Neurorehab Neural Res 1997; 11: 219–232.
- Xu D, Hong Y, Li J, Chan K. Effect of tai chi exercise on proprioception of ankle and knee joints in old people. Br J Sports Med 2004: 38: 50–54.
- Tseng YT, Tsai CL, Chen FC, Konczak J. Position sense dysfunction affects proximal and distal arm joints in children with developmental coordination disorder. J Mot Behav 2017; 28: 1–10.
- Tseng YT, Tsai CL, Chen FC, Konczak J. Wrist position sense acuity and its relation to motor dysfunction in children with developmental coordination disorder. Neurosci Lett 2018: 674: 106–111.
- Hall LA, McCloskey DI. Detections of movements imposed on finger, elbow and shoulder joints. J Physiol 1983; 335: 519–533.
- Refshauge K, Chan R, Taylor J, McCloskey D. Detection of movements imposed on human hip, knee, ankle and toe joints. J Physiol 1995; 488: 231–241.
- Wolpert DM, Pearson KG, Ghez C. The organization and planning of movement. In: Kandel ER, Schwartz JH, Jessell TM, Siegelbaum SA, Hudspeth AJ, editors. The principles of neural science. New York: McGraw Hill; 2013, p. 743–767.
- Blakemore SJ, Frith CD, Wolpert DM. The cerebellum is involved in predicting the sensory consequences of action. Neuroreport 2001; 12: 1879–1884.
- Chen FC, Wu SK. Perceived hole size, performance, and body movement during putting in children with and without probable developmental coordination disorder. Motor Control 2013; 17: 382–398.
- Chen FC, Tsai CL, Chang WD, Li YC, Chou CL, Wu SK. Postural control of anteroposterior and mediolateral sway in children with probable developmental coordination disorder. Pediatr Phys Ther 2015; 27: 328–335.
- 13. Henderson SE, Sugden DA. Movement assessment battery for children. 2nd edn. London: Harcourt; 2007.
- Conners CK. Conners' Rating Scale-Revised (CRS-R) technical manual. Toronto: Multi-Health Systems; 2001.
- 15. Kaufman AS, Kaufman NL. KBIT-2 Kaufman Brief Intelligence Test. 2nd edn. Minneapolis MN: NCS Pearson: 1997.
- van Melick N, Meddeler BM, Hoogeboom TJ, Nijhuis-van der Sanden MWG, van Cingel REH. How to determine leg dominance: the agreement between self-reported and observed performance in healthy adults. PLOS One 2017; 19: e0189876.
- Ghai S, Driller MW, Masters RSW. The influence of belowknee compression garments on knee-joint proprioception. Gait Posture 2018; 60: 258–261.
- 18. Payne K, Berg K, Latin RW. Ankle injuries and ankle strength, flexibility, and proprioception in college basketball players. J Athl Train 1997; 32: 221–225.
- Taylor RA, Marshall PH, Dunlap RD, Gable CD, Sizer PS. Knee position error detection in closed and open kinetic chain tasks during concurrent cognitive distraction. J Orthop Sports Phys Ther 1998; 28: 81–87.
- 20. Yokoyama S, Matsusaka N, Gamada K, Ozaki M, Shindo H. Position-specific deficit of joint position sense in ankles with chronic functional instability. J Sports Sci Med 2008; 7: 480–485.
- 21. Beynnon BD, Renstrom PA, Konradsen L, Elmqvist LG, Gottlieb D, Dirks M. Validation of techniques to measure

- knee proprioception. In: Lephart SM, Fu FH, editors. Proprioception and neuromuscular control in joint stability. Pittsburgh: Human Kinetics; 2000. p. 127–138.
- Konradsen L, Ravn JB, Sorensen AI. Proprioception at the ankle: the effect of anaesthetic blockade of ligament receptors. J Bone Joint Surg Br 1993; 75: 433–436.
- Ashton-Miller JA, Wojtys EM, Huston LJ, Fry-Welch D. Can proprioception really be improved by exercises? Knee Surg Sports Traumatol Arthrosc 2001; 9: 128–136.
- 24. Deshpande N, Connelly DM, Culham EG, Costigan PA. Reliability and validity of ankle proprioceptive measures. Arch Phys Med Rehabil 2003; 84: 883–889.
- Proske U, Gandevia SC. The proprioceptive senses: their roles in signaling body shape, body position and movement, and muscle force. Physiol Rev 2012; 92: 1651–1697.
- Hadders-Algra M. Developmental coordination disorder: is clumsy motor behavior caused by a lesion of the brain at early age. J Neural Transplant Plast 2003; 10: 39–50.
- Vaivre-Douret L, Lalanne C, Ingster-Moati I, Boddaert N, Cabrol D, Dufier JL, et al. Subtypes of developmental coordination disorder: research on their nature and etiology. Appl Neuropsychol Child 2011; 36: 614–643.
- 28. Chen FC, Li LL, Chu CH, Pan CY, Tsai CL. Finger soaking enhances effects of light touch on reducing body sway in children with developmental coordination disorder. J Rehabil Med 2019; 51: 217–224.
- Tseng YT, Holst-Wolf JM, Tsai CL, Chen FC, Konczak J. Haptic perception is altered in children with developmental coordination disorder. Neuropsychologia 2019; 127: 29–34.
- Artigas NR, Eltz GD, do Pinho AS, Torman VB, Hilbig A, Rieder CR. Evaluation of knee proprioception and factors related to Parkinson's disease. Neurosci J 2016: e6746010.
- 31. Teasdale H, Preston E, Waddington G. Proprioception of the ankle is impaired in people with Parkinson's disease.

- Mov Disord Clin Pract 2017; 4: 524-528.
- 32. McLeod KR, Langevin LM, Goodyear BG, Dewey D. Functional connectivity of neural motor networks is disrupted in children with developmental coordination disorder and attention-deficit/hyperactivity disorder. Neuroimage-Clin 2014; 4: 566–575.
- 33. Biotteau M, Chaix Y, Blais M, Tallet J, Péran P, Albaret J. Neural signature of DCD: a critical review of MRI neuroimaging studies. Front Neurol 2016; 7: 27.
- 34. Lincoln NB, Jackson JM, Adams SA. Reliability and revision of the Nottingham Sensory Assessment for stroke patients. Physiother 1998; 84: 358–365.
- 35. Stolk-Hornsveld F., Crow JL, Hendriks EP, van der Baan R, Harmeling-van der Wel BC. The Erasmus MC modifications to the (a revised) Nottingham Sensory Assessment: a reliable somatosensory assessment measure for patients with intracranial disorders. Clin Rehabil 2006; 20: 160–172.
- Lu L, Barrett A, Cibula J, Gilmore R, Heilman KM. Proprioception more impaired distally than proximally in subjects with hemispheric dysfunction. Neurol 2000; 55: 596–597.
- Madigan ML, Davidson BS, Nussbaum MA. Postural sway and joint kinematics during quiet standing are affected by lumbar extensor fatigue. Hum Movement Sci 2006; 25: 788-799.
- 38. Patel M, Fransson PA, Lush D, Gome S. The effect of foam surface properties on postural stability assessment while standing. Gait Posture 2008; 28: 649–656.
- Pal AR, Pratihar DK. Estimation of joint torque and power consumption during sit-to-stand motion of human-being using a genetic algorithm. Procedia Comput Sci 2016; 96: 1497–1506.
- Clark NC, Röijezon U, Treleaven J. Proprioception in musculoskeletal rehabilitation. Part 2: Clinical assessment and intervention. Man Ther 2015; 20: 378–387.

Appendix I. Position error data in each age group of children with DCD, and TD children

	PE at knee joint	PE at ankle joint
9-year-old children with DCD $(n=7)$	7.74 ± 5.66	9.65 ± 2.59
10-year-old children with DCD $(n=7)$	6.46 ± 3.40	9.25 ± 2.28
11-year-old children with DCD $(n=7)$	5.46 ± 1.25	8.10 ± 1.55
12-year-old children with DCD $(n=7)$	5.19 ± 1.96	8.07 ± 2.24
All children with DCD $(n=28)$	6.21 ± 3.45	8.77 ± 2.19
9-year-old TD children $(n=7)$	4.50 ± 1.02	4.32 ± 1.83
10-year-old TD children $(n=7)$	4.34 ± 0.66	3.73 ± 1.41
11-year-old TD children $(n=7)$	4.30 ± 1.33	3.70 ± 1.05
12-year-old TD children $(n=7)$	3.92 ± 1.26	3.15 ± 1.02
All TD children $(n=28)$	4.27 ± 1.06	3.73 ± 1.36
All participants (n = 56)	5.24 ± 2.71	6.25±3.12

DCD: developmental coordination disorder; TD: typically developing; PE: position error; PEV: position error variability.

Appendix II. Position error variability data in each age group of children with DCD, and TD children

	PEV at knee joint Children with DCD	Children with TD
9-year-old children with DCD $(n=7)$	3.64±2.71	4.69 ± 1.10
10-year-old children with DCD $(n=7)$	2.93±1.68	4.39 ± 1.08
11-year-old children with DCD $(n=7)$	2.59±0.59	4.23 ± 100
12-year-old children with DCD $(n=7)$	2.51±0.93	3.93 ± 0.93
All children with DCD $(n=28)$	2.92±1.65	4.31 ± 1.01
9-year-old TD children (n = 7)	2.13 ± 0.49	2.19 ± 0.75
10-year-old TD children (n = 7)	2.06 ± 0.31	1.97 ± 0.83
11-year-old TD children (n = 7)	2.04 ± 0.63	1.87 ± 0.92
12-year-old TD children (n = 7)	1.86 ± 0.6	1.63 ± 0.68
All TD children (n = 28)	2.02 ± 0.50	1.91 ± 0.78
All participants (n = 56)	2.47±1.29	3.11 ± 1.50

DCD: developmental coordination disorder; TD: typically developing; PE: position error; PEV: position error variability.

Appendix III. Correlation between proprioceptive acuity data and subscore in children with DCD, and TD children

	Children with DCD		Children with TD			
	Manual dexterity	Aiming & catching	Balance	Manual dexterity	Aiming & catching Balance	
PE at knee joint	r = 0.05, p=0.98	r=0.32, p=0.10	r=-0.47, p=0.01*	r=-0.02, p=0.93	r=-0.05, p=0.81	r=-0.11, p=0.57
PE at ankle joint	r = -0.21, p = 0.29	r = 0.19, p = 0.34	r=-0.41, $p=0.03*$	r = -0.10, p = 0.61	r = -0.12, p = 0.53	r = 0.05, p = 0.79
PEV at knee joint	r = 0.03, p = 0.89	r = 0.34, p = 0.08	r = -0.44, p = 0.02*	r = -0.17, p = 0.93	r = -0.05, $p = 0.81$	r = -0.11, p = 0.56
PEV at ankle joint	r = -0.20, p = 0.32	r = -0.04, $p = 0.85$	r = -0.42, p = 0.03*	r = 0.18, p = 0.93	r = -0.06, $p = 0.77$	r = -0.03, $p = 0.87$

DCD: developmental coordination disorder; TD: typically developing; PE: position error; PEV: position error variability.