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## **Joint-Position Sense and Kinesthesia in Cerebral Palsy**

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## **Abstract**

**Objectives:** Examine joint-position sense and kinesthesia in all extremities in participants with diplegic or hemiplegic cerebral palsy (CP).

**Design:** Survey of joint-position sense and kinesthesia differences between aged-matched controls and 2 groups with CP.

**Setting:** University movement assessment laboratory.

**Participants:** Population-based sample of participants with CP, diplegia (n=21), hemiplegia  $(n=17)$ , and age-matched volunteers  $(n=21)$  without neurologic disease.

**Interventions:** Not applicable.

**Main Outcome Measures:** Joint-position sense and kinesthesia were measured in the transverse plane (forearm pronation/supination and hip internal/external rotation) using a custom built device. For joint-position sense, participants actively rotated the tested limb to align the distal end with 10 target positions first with the limb and targets visible to assess their ability to perform the task motorically. The task was then repeated with vision of the limb occluded, with targets remaining visible. Joint-position sense error was determined by the magnitude and direction of the rotation errors for each limb in the vision and no vision conditions. Kinesthesia was evaluated by the ability to detect passive limb rotation without vision.

**Results:** No group differences were detected in the vision condition. Indicative of joint-position sense deficits, a significant increase in errors was found in the no vision condition in all limbs except the dominant upper limb for both groups with CP. Joint-position sense errors were systematically biased toward the direction of internal rotation. Kinesthesia deficits were evident on the nondominant upper limb in diplegia and hemiplegia, and bilaterally in the lower limbs in hemiplegia. In hemiplegia, joint-position sense and kinesthesia deficits were noted on the dominant limbs, but were significantly worse on the nondominant limbs.

**Conclusions:** These results indicate that people with CP have proprioception deficits in all limbs.

#### **Keywords**

Cerebral palsy; Proprioception; Somatosensory; Rehabilitation

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Proprioception is a complex somatosensory modality that utilizes inputs from muscle, joint, and cutaneous afferent fibers, and consists of 2 components, the sense of limb movement (kinesthesia) and static limb position (joint-position sense).<sup>1</sup> People with CP have well  $d$  documented balance impairments<sup>2</sup> and tend to rely disproportionately on visual input to maintain posture and to position their limbs during gait, which may reflect deficits in proprioception.<sup>3</sup> Few studies have examined proprioception in CP, and none have differentiated and examined both joint-position sense and kinesthesia. Most studies evaluated proprioception in participants with hemiplegia,  $\frac{4}{3}$  some reported combined data from various subtypes and severities of CP, and few examined proprioception in those with diplegia, despite similar prevalence of both clinical conditions.<sup>5</sup> Consequently, it is difficult to associate specific deficits with different subtypes of CP or to distinguish between effects on joint-position sense compared to kinesthesia. Methods used in prior studies of proprioception in CP primarily were clinically-based, typically yielded nominal data, and often did not include a control or comparison group. Only 1 study assessed joint-position sense, where participants matched elbow joint position to a remembered position.<sup>6</sup> Kinesthesia, studied to a greater extent than joint-position sense, has typically been assessed by having participants determine the direction of a passively moved limb (eg, "Did your toe move up or down?") without controlling for force of cutaneous contact or limb-movement displacement and velocity.<sup>4,7-15</sup> In hemiplegia, kinesthesia impairments on the nondominant upper limb have been demonstrated consistently, most often in comparison to the contralateral dominant limb, with limited data describing kinesthesia on the dominant or nondominant limbs compared to controls.<sup>7,12,15</sup> Some studies of diplegia found upper-limb kinesthesia deficits,  $8,16$  but others noted largely intact kinesthesia in these limbs.10 Most studies in CP assessed kinesthesia of the index finger or elbow, and only 2 studies reported significant deficits in lower-limb joint proprioception in  $diplegia.$ <sup>10,11</sup>

We assessed joint-position and kinesthesia in the upper and lower limbs bilaterally using a more sensitive parametric testing protocol in participants with hemiplegic and diplegic CP and an age-matched group without disability. Joint-position sense and kinesthesia were measured in the transverse plane (forearm pronation/supination and hip internal/external rotation) where rotational abnormalities are commonly seen. No prior study in this population investigated proprioception in the transverse plane or of large joints such as the hips. We hypothesized that participants with CP would show diminished joint-position sense and kinesthesia in their more impaired limbs (eg, the lower limbs in diplegia and the side of involvement in hemiplegia) compared to controls, and would demonstrate abnormal but less severely diminished proprioception in unaffected or less impaired limbs.

## **METHODS**

#### **Partcipants**

Fifty-nine people participated in this study. The sample included 21 participants with spastic diplegia (mean age  $\pm$  SD, 14y10mo $\pm$ 7y; age range, 7y4mo–34y3mo; 10 males), 17 participants with hemiplegia (mean age  $\pm$  SD, 13y11mo $\pm$ 5y6mo; age range, 8y mo–26y6mo; 6 males), and 21 age-matched control subjects (mean age ± SD, 14y10mo±SD5y1mo; age range, 7y6mo– 24y4mo; 11 males) without neurologic or orthopedic disabilities. All participants ambulated independently and participants with CP were either Level I or II on the  $GMFCS<sup>17</sup>$  and MACS. <sup>18</sup> Additionally, school-age participants were in grade-levels appropriate within 2 years of their age; all those over 21 years were in or had completed college; and all participants reliably followed instructions.

All participants aged 17 years or younger provided informed assent following guidelines approved by the Human Studies Committee of Washington University, and their parent or legal guardian provided informed consent; all participants aged 18 years or older provided informed

consent. Responses to a modified Edinburgh Handedness Inventory\*19 indicated the proportion of right upper-extremity dominance for each participant (a score of 100 indicates complete right upper-extremity dominance, and a score of 0 indicates complete left upperextremity dominance). Leg dominance was assumed to be ipsilateral to upper-extremity dominance.

#### **Proprioception Protocol**

A custom built device allowed rotation around the axis of a semi-goniometer and thereby measured hip and forearm orientation angles in the transverse plane (fig 1), more specifically hip internal and external rotation and forearm pronation and supination. During testing, the foot and lower calf or hand and forearm were placed in a foam lined holder that accommodated variable limb sizes and biomechanical alignment differences with adjustable padding and Velcro straps. Tactile cues were minimized with secure positioning and smooth axial rotation through a square mounted-flanged ball bearing.

#### **Joint-Position Sense**

Joint-position sense was assessed by having participants actively point with a specified part of their foot or hand to each of 10 target angles along the semi-goniometer axis. These target angles were randomly selected prior to the beginning of the experiment. All participants received the same 10 target angles, presented in a random order without replacement. The range of excursion within which the target angles were located was predetermined through pilot testing to be within the available active range for most participants with mild CP (ie,  $60^{\circ}$  arc for the forearm and 35° for the hip). Targets were large numbers at every 10° increment or hash marks at every degree located on both sides of the goniometer and were plainly visible to all participants (see fig 1). The target number was both stated and pointed to by the tester. The orientation angle of the leg or arm was approximated to the nearest degree; the measurement error for approximation of the limb was not directly assessed. A single tester completed all assessments.

Testing of upper and lower extremities involved 2 conditions. In the vision condition, participants saw both target location and their foot or hand; in the no vision condition the limb was obscured by an opaque curtain and only targets on the goniometer were visible, requiring somatosensory input for limb guidance to complete the task. Participants performed the vision condition first to facilitate instruction and assess their motor abilities with respect to performing the task. The vision condition was followed by the no vision condition to assess joint-position sense impairments.

When testing forearm rotation, participants sat upright at a table directly in front of the goniometer. The tested arm was unrestrained and positioned comfortably in approximately 90° of elbow flexion. In the vision condition for the upper extremity, the pointing reference used to align with the target was the center of the padded handle, which was gripped lightly by the participant and normative to the axis of rotation (fig 1A). When testing the leg, participants sat in a semi-reclined position with the back supported, a hip angle of approximately 45° of flexion, and knee in full extension. In the vision condition, the pointing reference used to align with the target was a small marker placed on the dorsal surface of the midline of the second toe (fig 1B). In the no vision condition, target alignment was made without sight of the pointing reference and associated limb which was covered by a curtain. For each limb and both conditions, the magnitude (degrees) and direction (internal/external rotation or pronation/

 $^*$ The modified Edinburgh Handedness Inventory determines the proportion of upper-extremity usage during the following tasks: writing, drawing, throwing, scissoring, brushing teeth, knife and spoon use, upper hand on broom, striking match, and opening a box lid.

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supination) of error between performance and target location were recorded for each trial to the nearest degree.

#### **Kinesthesia**

The same goniometer and limb positioning device was used to evaluate kinesthesia, but vision of the limbs was obscured during all trials. Participants immediately reported movement direction upon detecting passive rotation of approximately 0.5°/s with a maximum displacement of 4°. Movement was imposed passively using a control rod attached to the back of the goniometer. Direction was pseudorandomly selected per trial, with half of the trials moving in one direction and half in the other direction. Performance accuracy was the number of correct responses in 10 trials for each limb.

#### **Statistical Analyses**

Post hoc 1-tailed Mann-Whitney *U*† tests examined pair-wise differences within and between groups for the joint-position sense and kinesthesia tasks. Groups with diplegia and hemiplegia were each compared to controls but not to each other. Mann-Whitney *U P* values were Bonferroni corrected for multiple comparisons on the basis of 6 comparisons per task.

## **RESULTS**

Deficits in joint-position sense and kinesthesia were found in both subtypes of CP, most notably on the nondominant limbs. Within and between group differences are described separately for groups with diplegia and hemiplegia.

#### **Diplegia Compared to Controls**

The group with diplegia and the control group had comparable joint-position sense error distributions in all limbs when allowed to use vision (fig 2) (table 1C: see columns CD-DD and CN-DN). Within-group comparisons for both diplegia and controls in the vision condition revealed that the nondominant lower extremity had significantly larger mean joint-position sense error compared to the dominant side (fig 2B). Comparable performance in the vision trials between groups critically indicates that the motor disability in diplegia did not impair performance on this task with any limb. In contrast, with exclusive reliance on somatosensory input in the 'no vision' trials, the group with diplegia made significantly larger joint-position sense errors than controls in both lower limbs (fig 2B) (table 1D: see columns CD-DD and CN-DN) and the nondominant upper limb (fig 2A) (table 1D: see column CN-DN). Additionally, the distribution and mean joint-position error in those with diplegia was biased internally, in the direction of pronation for the nondominant forearm and hip internal rotation for both legs (see fig 2). In the no vision trials with the dominant upper limb, errors were larger, but not significantly, in diplegia compared to controls (see fig 2A) (table 1D: see column CD-DD). Compared to the vision condition, the group with diplegia had significantly greater jointposition sense errors in the no vision condition in the nondominant upper limb (mean difference, −2.11±8.33°, *P*=.006) and both lower limbs (dominant: mean difference, −1.53 ±8.47°, *P*<.001; nondominant: mean difference, −5.24±10.47°, *P*<.001). For the control group, only the nondominant lower limb had significantly larger errors across conditions (mean difference, −1.27±°, *P*<.001). Within diplegia both nondominant limbs had significantly larger joint-position sense error than their respective dominant limbs in the no vision condition (see fig 2) (table 1D: see column DD-DN).

<sup>†</sup>The Mann-Whitney *U* test was used because variances were unequal across groups.

On the kinesthesia test, the group with diplegia detected passive movements of all limbs less accurately than controls (fig 3) (table 2A: see columns CD, CN, DD, DN; and table 2B: see columns CD-DD and CN-DN). However, this difference was significant only for both upper limbs (fig 3A) (table 2B: see columns CD-DD and CN-DN), and approached significance for the nondominant lower limb (see table 2B: see column CN-DN). Absence of a significant difference, especially in the nondominant lower limb, possibly reflected greater variation across participants with diplegia, with the performance of some participants overlapping that of controls.

#### **Hemiplegia Compared to Controls**

The group with hemiplegia and the control group had comparable joint-position error distributions in all limbs when allowed to use vision (fig 2) (table 1C: see columns CD-HD and CN-HN). Comparable group performance in the vision condition indicates that movement disabilities in hemiplegia did not contribute to proprioception deficits. In contrast, with exclusive reliance on somatosensory input in the no vision trials (fig 2B), the group with hemiplegia made significantly larger joint-position sense errors than controls with both lower limbs (table 1D: see columns CD-HD and CN-HN). For both upper limbs, mean errors in hemiplegia were larger than in controls in the no vision trials (see fig 2A), although these differences were not significant. Similar to the diplegia group, the distribution and mean jointposition error was in the direction of pronation for the nondominant forearm and hip internal rotation for both legs (see fig 2). Additionally, the group with hemiplegia had significantly greater joint-position sense errors in the no vision condition compared to the vision condition in both lower limbs (dominant: mean difference, −4.69±13.80°, *P*<.001; nondominant: mean difference, −5.68±10.89°, *P*<.001), and this difference approached significance in the nondominant upper limb (mean difference, −2.55±9.95°, *P*<.059).

The group with hemiplegia performed the kinesthesia task significantly less accurately than controls with the nondominant arm and both legs (fig 3) (table 2B: see columns CD-HD and CN-HN). Mean accuracy was diminished, though not significantly, even with the dominant forearm (fig 3A). However, this difference was not significant, likely the result of greater variability in accuracy in hemiplegia.

Subsequent post hoc Mann-Whitney *U* tests assessed performance accuracy differences between older and younger participants in each group. Mann-Whitney *U* tests found no effect of age after stratification by age into groups less than 13 years and 13 years and older.

## **DISCUSSION**

Pervasive proprioception deficits in all limbs except the dominant upper extremity were found in participants with diplegia or hemiplegia with relatively mild motor involvement (GMFCS and MACS Levels I or II). The nondominant upper limb in both groups with CP had a jointposition sense error greater than double the error magnitude of controls, and both lower limbs in CP had more than a 3-fold greater error magnitude compared to controls (see table 1). Significant proprioception deficits were observed in those limbs that previously might be considered less involved such as the upper extremities of participants with diplegia and the dominant upper and lower extremities of those with hemiplegia. A recent study of participants with mild diplegia or hemiplegia also exhibited tactile sensory deficits in both hands.<sup>20</sup> Taken together, the distribution of tactile deficits and the proprioception deficits shown here indicate that CP involves generalized somatosensory deficits. Such behavioral evidence of generalized somatosensory deficits corroborate recent diffusion tensor imaging in participants with diplegic CP showing severely damaged thalamocortical projections to the somatosensory cortex, with less frequent damage to the corticospinal tracts.<sup>21</sup> Therefore, the proprioception deficits observed here most probably result from primary central nervous system lesions in CP

that affect all known proprioceptive inputs to the cortex arising from muscle spindles, Golgi tendon organs, and the array of sensory afferent innervation of the joints and skin. Unknown, however, is the extent to which secondary effects related to decreased or abnormal limb use also contribute to somatosensory deficits. For example, muscle spindles, potentially altered from spasticity or muscle-tendon shortening, may contribute to proprioception errors, especially the bias toward internal errors noted here, since spastic muscle fibers have been shown to be stiffer and sarcomeres shorter than normal.<sup>22</sup> Further research directed at determining the precise etiology of proprioception deficits and the systematic directional bias of errors needs to be conducted. We analyzed proprioception deficits with respect to limb dominance as defined from responses to a modified Edinburgh Handedness Inventory<sup>19</sup> for upper-extremity dominance and then projected this categorization to the ipsilateral leg. We are aware of no similar validated assessment for leg dominance. Additionally, as part of the Edinburgh Handedness Inventory participants were asked to name the leg they typically use for kicking, which has been used as an indicator of leg dominance in those without disability, and which always corresponded to the side of hand dominance in our sample. However, the act of kicking with 1 leg requires balancing on the opposite leg, so balance difficulties in CP could lead to balancing on the more functional limb and kicking with the more affected leg. Nonetheless, any potential incorrect categorization of lower-limb dominance did not compromise these findings because significant proprioception deficits were observed bilaterally in both patient groups. Based on observing proprioception deficits in each limb, the following discussion considers the implications of these findings for the upper and lower extremities separately.

#### **Upper-Extremity Proprioception Deficits in CP**

Proprioceptive deficits in the upper limbs of participants with CP were greater on the nondominant side. On the nondominant arm in diplegia and hemiplegia, selected joint-position sense errors were significantly larger, especially in pronation, and passive movements were significantly less accurately detected. Our assessments of proprioception deficits on the nondominant arm in hemiplegia are similar to those previously reported.<sup>4,8,12-15</sup>

With 1 exception,  $\frac{4}{7}$  prior studies of proprioception did not report deficits in the dominant arm in hemiplegia.<sup>7,12-16</sup> No significant dominant arm impairments for joint-position sense or detection of passive movements were found in our sample of participants with hemiplegia. In the kinesthesia task, however, mean accuracy was lower and the minimum-to-maximum range was larger even for the dominant arm in hemiplegia compared to controls. Inclusion of participants with greater motor impairment (higher GMFCS level) might reveal greater dominant arm deficits in proprioception.

#### **Lower-Extremity Proprioception Deficits in CP**

Proprioception impairment in the lower extremities can directly impact balance and gait.<sup>23</sup> Bilateral joint-position sense deficits were found in the lower extremities in both patient groups. Due to the inclusion of the vision condition, we ascertained that differences in motor performance, in the groups with CP, were not the source of the observed proprioception deficits. Previous studies used simple detection of small, passive sagittal plane movements of the index finger, great toe, and knee to reveal proprioception deficits in diplegia,10,11 but no studies described bilateral lower-extremity deficiencies in hemiplegia. In the present study, both groups had significantly increased error in rotating to target angles when vision was occluded compared to controls. Consistent with previous findings that visual input is necessary for gross motor function in  $\mathbb{CP}^3$  comparable errors were not observed when these participants were able to see the leg and targets simultaneously. These findings suggest that participants with CP utilize visual input as a compensatory mechanism for activities involving joint-position sense. Normalized motor control depends on visual, vestibular, and proprioceptive inputs. However, during visuomotor tasks, vision predominates over proprioception, especially when the 2 sources of information conflict.<sup>24</sup>

Lower-extremity proprioception was assessed in the current study in a nonweight-bearing condition. Proprioceptive deficits similar to those observed here are also likely to exist in weight-bearing activities such as standing and gait since perception of a limb's position and/ or movement in space is independent of body position and input from load bearing. Somatosensory feedback during weight bearing activities includes multiple additional sources of afferent input, including tactile and pressure receptors. While these possibly obscure or compensate for proprioceptive deficits, they may also be similarly affected given evidence of widespread tactile impairments in the upper extremities in people with  $CP<sup>20</sup>$  Future evaluation of somatosensation during weight bearing is a critical component to understanding the role of somatosensory deficits in the motor disorders related to CP.

The primary directional bias of errors when detecting joint-position in both patient groups was towards internal hip rotation. Prevalent internal rotation errors possibly reflect common lower extremity musculoskeletal alignment in participants with CP, which often includes increased internal femoral torsion and hip adduction during standing and walking compared to those without disabilities. Possibly abnormal biomechanical alignment, muscle weakness or imbalance, and/or increased muscle tone related to CP biases proprioceptive input towards internally oriented joint-position errors. Secondary muscle changes related to spasticity also might impair joint-position sense over time by shortening and stiffening muscle tissue,  $22,25$ altering the muscle-joint relationship<sup>26</sup> and disrupting the sensitivity of muscle spindles, which contribute to proprioception.23 Unknown, however, is whether the extent of effects on proprioception receptors in CP is secondary to disrupted corticofugal influence on motor behavior due to injured thalamocortic projections. Thus, central nervous system somatosensory deficits might contribute to or even precede abnormal biomechanical alignment in CP. Alternatively, centrally precipitated alterations in muscle physiology plausibly cascade with secondary changes in the sensitivity of the panoply of proprioception peripheral afferents. Lower accuracy on the joint-position compared to the kinesthesia task possibly arose from task differences in difficulty or in the amount of tactile information provided. Plausibly tactile stimulation was reduced when judging a static joint-position compared to coincident stimulation when the examiner passively moved the limb during the kinesthesia task. Tactile stimulation was minimized here by using custom fitted foam liners around the foot. However, detecting the direction of movement in the kinesthesia task was possibly easier due to retained extraneous tactile cues. Additionally, the joint-position task probably was more difficult because it involved active angular rotations that had to be precisely graded. In contrast, the dichotomous kinesthesia task required less precision when detecting external or internal rotations that were passively imposed. Another possibility for smaller group accuracy differences in the kinesthesia compared to joint-position sense task is a smaller effect size on the kinesthesia task, which necessitated a larger sample size to indicate group differences in individual limbs.

#### **CONCLUSIONS**

#### **Study Limitations**

This study minimized confounding effects from movement limitations by studying participants with CP who have milder impairment and by comparing performance within participants on the active positioning task with and without the use of vision. This study additionally advances the study of proprioception over prior protocols by introducing a novel method of joint-position sense assessment and improving kinesthesia assessment by controlling the magnitude and area of skin contact and movement velocity and displacement.

The definitive proprioception deficits found here in participants with mild CP are in contrast to prior inconsistent findings that primarily utilized routine clinical tests. Current findings indicate the need to assess proprioception separately for each limb and to evaluate results against comparably tested, age-matched controls. Furthermore, evaluations based on bilateral limb position matching-tasks are probably incomplete because deficits are often bilateral in CP, especially in the lower extremities. More accurate assessment of how participants perceive limb position and movement can be obtained by testing limbs separately. Revealed somatosensory deficits might thereby provide rehabilitation clinicians with information about factors contributing to motor impairments. Indeed, even limbs less motorically affected (eg, upper limbs in diplegia or less affected lower limb in hemiplegia) showed impaired proprioception.

Improved performance accuracy when seeing the affected limb during the tasks revealed a probable compensatory, visual adaptation to pervasive proprioception deficits in CP. Therefore, optimization of vision is essential for people with CP. Participants' vision should be engaged and relied upon while learning and practicing movements. Reliance on visual compensation strategies (eg, using mirrors, video, virtual reality) is important early in the rehabilitation process until accurate perception of body movements is demonstrated. Subsequently, additional benefits might arise from having participants practice motor tasks with gradually decreasing visual input such that even diminished proprioception can be incorporated into learned movements.27 Practicing movements and proprioception tasks without vision also might improve perception as a neural recovery strategy by repeatedly engaging the somatosensory system during the activity.<sup>28</sup>

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### **List of Abbreviations**

CP, cerebral palsy; GMFCS, Gross Motor Function Classification System; MACS, Manual Ability Classification System; CD, Control group, dominant side; CN, Control group, nondominant side; DD, Diplegia group, dominant side; DN, Diplegia group, non-dominant side; HD, Hemiplegia group, dominant side; HN, Hemiplegia group, non-dominant side.

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#### **Fig 2.**

Box-and-whisker plots of group magnitude (in degrees) and direction of joint-position sense error. Ten targets were used for each limb and participant. The 4 plots indicate findings for upper (A) and lower (B), nondominant and dominant limbs, for controls (black), diplegia (grey), and hemiplegia (white) on the vision and no vision condition trials. The vertical green bars within the boxes mark group medians and the red crosses mark group means. The boxes represent the first and third quartile of the data and the error bars represent the 95% confidence intervals around the median. The negative abscissa represents pronation for arms and hip internal rotation for legs and the positive represents supination and hip external rotation. The *P* value is shown where group differences were significant.

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**B. Lower Extremity** 



#### **Fig 3.**

Bar graphs of accuracy (mean and SD) in detecting passive limb movements by group for upper (A) and lower (B) and for dominant (filled) and nondominant (cross-hatched) limbs in controls (black), diplegia (grey), and hemiplegia (white). \* *P*<.05.



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 NIH-PA Author ManuscriptNIH-PA Author Manuscript **Table 2**



NOTE. Mann-Whitney UP values were adjusted with the Bonferroni correction for 6 comparisons, and are shaded where differences between group means were significant. Abbreviations: CD, control dominant side; CV, control nond NOTE. Mann-Whitney *U P* values were adjusted with the Bonferroni correction for 6 comparisons, and are shaded where differences between group means were significant. Abbreviations: CD, control dominant side; CN, control nondominant side; DD, diplegia dominant side; DN, diplegia nondominant side; HD, hemiplegia dominant side; HN, hemiplegia nondominant side; L, lower extremity; U, upper extremity.