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Relationship between kinematic motion abnormalities and bimanual performance in children with unilateral cerebral palsy

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ABSTRACT

AIM: To evaluate the relationship between the movement abnormalities of the impaired upper limb in children with unilateral cerebral palsy (CP) and bimanual performance.

METHOD: Twenty-three children with unilateral CP (mean age 11y 10mo [SD 2y 8mo]) underwent evaluation of bimanual performance (Assisting Hand Assessment [AHA]) and a three-dimensional movement analysis to measure deviations in the movement of their affected upper limb, and compared with 23 typically developing children (TDC) (mean age 11y 11mo [SD 2y 5mo]). Kinematic indices, such as the Global Arm Profile Score (APS), which summarizes the global movement deviation of the upper limb from the norm, and the Global Arm Variable Score (AVS), which represent movement deviations for a given joint, were calculated and correlated to AHA.

RESULTS: Values of kinematic indices were significantly higher in children with unilateral CP than in TDC. A strong correlation between Global-APS and AHA score ($r = 0.75$) was found. Other significant correlations were found with Global-AVS, especially in distal joints.

INTERPRETATION: Children with unilateral CP had higher movement deviations than TDC. The global movement deviation of the impaired upper limb was strongly correlated with bimanual performance. The influence of distal abnormalities confirms the importance of considering these limitations in therapeutics.

WHAT THIS PAPER ADDS

- Children with unilateral cerebral palsy had higher movement deviations than typically developing children in unimanual tasks.
- A strong relationship was found between movement deviations of the impaired upper limb and bimanual performance.

ABBREVIATIONS

3DMA : Three-Dimensional Movement Analysis

AHA : Assisting Hand Assessment

APS : Arm Profile Score

AVS : Arm Variable Score

CP : Cerebral Palsy

MACS : Manual Ability Classification Scale

TDC : Typically Developing Children

INTRODUCTION

Children with unilateral Cerebral Palsy (CP) have variable upper limb impairments, including spasticity, sensorimotor deficits¹, synkinetic movements², lack of selective motor control and difficulties in motor planning³.

For these children, hand function is especially affected during bimanual tasks^{4,5} because it requires motor coordination between the two hands. Understanding of the bimanual performance, i.e how the child spontaneously uses his or her impaired limb in bimanual tasks⁶, is important to better target clinical treatments. Previous studies showed the impact of different parameters such as tactile function⁷ or grip strength⁸ on bimanual performance, but the influence of the movement deviations of the impaired upper limb has been poorly described and needs to be better understood.

The impaired motor control of the upper limb can be objectively measured using three-dimensional movement analysis (3DMA). Previous studies⁹⁻¹¹ found that children with unilateral CP have abnormal shoulder abduction and rotation, less elbow extension, less supination and more wrist flexion on their impaired limb than typically developing children (TDC). However, most of them are based on discrete kinematic parameters such as maximum joint angle or range of motion. They do not propose summarized kinematic indices that take into account the waveform of the movement and reflect the movement pattern. Only a few studies have proposed such kinematic indices for the upper limb^{12,13}, all based on unilateral movement. Those developed by Jaspers et al.¹² are certainly the most consistent because they summarize the severity of the upper limb movement pathology in a set of seven unilateral tasks by suggesting both global and segmental kinematic indices, with excellent psychometric properties in terms of reliability and validity.

The aim of the present study was to determine the relationship between the movement pathology of the impaired upper limb in children with unilateral CP, using 3DMA indices as suggested by Jaspers et al.¹², and the bimanual performance, evaluated with the Assisting Hand Assessment (AHA)¹⁴.

METHODOLOGY

This was an ancillary study, which is part of a larger clinical trial (NCT02131909) assessing the effect of mirror therapy in children with unilateral CP. Written agreement from the Committee to Protect People was obtained. Written consent of children and parents was obtained before the start of the study.

PARTICIPANTS

Children (aged 7-18 years old) with unilateral CP were recruited for the study at the Rennes University Hospital between March 2014 and July 2015. Eligible patients had a Manual Ability Classification Scale (MACS) level¹⁵ of I (handles objects easily) to III (handles objects with difficulty). Exclusion criteria were: upper-limb pain, severe cognitive or visual disturbances, upper-limb surgery or use of botulinum toxins within six months prior to the study. All assessments were performed on the day of inclusion. In addition, a control group of TDC were recruited and underwent the 3DMA protocol with their non-dominant limb.

The calculation of sample size was based on prior studies that observed bivariate correlations of at least $r = .67$ between kinematic indices and bimanual performance in unilateral CP patients. A statistical power analysis revealed that a sample size of at least 22 participants was required to replicate this effect with 95% power. Finally, 23 children with unilateral CP completed the study (14 boys / 9 girls, 14 left / 9 right hemiplegia, mean age = 11 years 10 months, standard deviation [SD] = 2 years 8 months). Three were classified as MACS I (*handles objects easily*), seventeen as MACS II

(*handles most objects*) and three as MACS III (*handles objects with difficulty*). The mean AHA score was 62.9 AHA-units (SD 11.3).

Concurrently, 23 TDC (11 boys / 12 girls, 22 right-handed / 1 left-handed, mean age = 11 years 11 months, SD = 2 years 5 months) were included in the study as a control group. (A total of 28 TDC completed the study; 5 participants were randomly excluded to balance sample size with the unilateral CP group.) The TDC sample was comparable to the unilateral CP sample in terms of sex ($p = 0.375$) and age ($p = 0.954$).

BIMANUAL PERFORMANCE

Bimanual performance was evaluated with the Assisting Hand Assessment (AHA)¹⁴. This tool has excellent psychometric properties in terms of validity, reliability and sensitivity to change¹⁴. Individual items assess general use, arm use, grasp-release, fine motor adjustments, coordination, and pace. The raw score and logits are converted on a 0-100 AHA-unit scale, where 100 AHA-units corresponds to an optimal spontaneous use of the assisting hand.

UPPER-LIMB KINEMATIC ANALYSIS

3DMA Protocol

3DMA data of the upper-limb motion were collected with 16 optoelectronic cameras at 100 Hertz (Optitrack, Corvallis, Oregon, USA). A 13-marker set, applied at trunk, arm and forearm, according to the recommendations of the International Society of Biomechanics (ISB)¹⁶ was used (see Figure S1, online supporting information).

The set of tasks performed by each child was derived from the motion analysis protocol designed by Jaspers et al., which has demonstrated its good validity and reliability in TDC¹⁷ and children with unilateral CP¹⁸. Seven unimanual tasks of this protocol were used, including two reach tasks (reach forwards until shoulder flexed at 90° and reach side-ways with shoulder abducted at 90° with palm facing the ground), three gross motor tasks (bring hand to mouth, to head and to contralateral shoulder) and two reach-to-grasp tasks (reach-to-grasp a glass vertically and a reach-to-grasp a ball horizontally). The children performed the tasks at self-comfortable speed. They were seated on a height-adjustable chair. For the reach-to-grasp tasks, the object was positioned on an adjustable table at arm's length. In the starting position, the child was seated, knees and hips flexed at 90°, and back straight. Contrary to the protocol of Jaspers et al., the start position of the impaired/non-dominant arm was not on the ipsilateral knee but alongside the body to ensure the best spontaneous position at the start.

Calculation of kinematic indices

The kinematics of the trunk (flexion, lateral flexion and rotation), elbow (flexion and pronation) and wrist (flexion and deviation) were calculated using the Euler sequences recommended by the International Society of Biomechanics¹⁶. The kinematic of the shoulder was assessed by calculating the thoracohumeral joint (abduction, flexion and rotation), using the XZY sequence⁽¹⁹⁾. The kinematic data were processed with Matlab (MathWorks, Natick, MA, USA).

For each task, three series of four movement cycles were performed (total of twelve movement cycles per task). A movement cycle began when the hand left the rest position and ended when the task was completed ("hand stop" for reach tasks and gross motor tasks and "beginning of the movement of a marker placed on the object" for the reach-to-grasp tasks). Movement cycles were time-normalized (0-100%).

Four types of kinematic indices were calculated, as described by Jaspers et al.¹² (see supplementary material II):

(1) The *Arm Variable Score* (AVS) is an index of deviation for a single joint angle during one particular task, calculated separately for the 10 joint movements. Each AVS was calculated as the root mean square error average between the three selected curves of one child for a given joint angle and the mean curve of the TDC group for the same joint angle.

(2) The *Arm Profile Score* (APS) reflects the total deviation of the upper limb during one particular task. It was calculated as the root mean square error average of the 10 joint movements during this task, separately for the seven tasks.

(3) The *Global-AVS* represents the deviation for a single joint angle among all seven tasks. It was calculated by averaging the seven AVSs of the joint considered.

(4) The *Global-APS* represents the global deviation of the upper limb in all the seven tasks and all 10 joint movements. It was calculated by averaging the seven APSs.

These kinematic indices, expressed in degrees, were calculated for the children with unilateral CP and the TDC. For each child, they reflect the average difference across time between his/her three own selected movement cycles and the average movement cycle of the TDC group. In sum, these kinematic indices summarize and grade the severity of the impaired/non-dominant upper limb movement deviation during a unimanual task. A higher value reflects a greater deviation from the database of TDC.

STATISTICAL ANALYSES

All statistical tests were performed with a two-tailed significance level of 5%. Multiple kinematic indices of the unilateral CP and TDC groups had an unequal variance and a non-normal distribution with right-side asymmetry, especially in the unilateral CP group. To ensure that this did not bias the results, all analyses were replicated using non-parametric tests (Mann–Whitney U test): the results were identical in all cases.

Firstly, the comparison between the unilateral CP and TDC groups on the kinematic indices was investigated by a two-way mixed-design analysis of variance (ANOVA; with group as a between-subject factor and kinematic index as a within-subject factor), separately for Global-AVS and APS scores. The unilateral CP and TDC groups were then compared on each kinematic index using Welch's unequal variances *t*-tests¹⁹.

Secondly, the relationships between kinematic indices and AHA scores were assessed using Pearson's correlation coefficient. A correlation coefficient greater than 0.70 was considered as high, 0.60 to 0.69 as good, 0.50 to 0.59 as moderate, 0.40 to 0.49 as fair, and less than 0.40 as weak²⁰.

RESULTS

COMPARISON OF KINEMATIC INDICES BETWEEN CHILDREN WITH UNILATERAL CEREBRAL PALSY AND TYPICALLY DEVELOPING CHILDREN

Concerning the Global-AVS, a two-way ANOVA revealed a significant main effect of group ($F[1,44] = 48.58$, $p < 0.001$, $\eta^2_p = 0.52$), indicating that the unilateral CP group demonstrated significantly larger deviation overall. The two-way interaction between group and Global-AVS index was also significant ($F[9,396] = 15.49$, $p < 0.001$, $\eta^2_p = 0.26$), suggesting that the difference between the two groups varied in magnitude across indices. As shown in Figure 1, the main effect of group, using *t*-tests, revealed that children with unilateral CP demonstrated significantly larger deviations than controls for all indices, except for trunk flexion and rotation. Deviations were more pronounced for more distal joints (elbow and wrist).

Concerning the APS, a two-way ANOVA revealed a significant main effect of group indicating that children with unilateral CP had overall larger deviations from the norm than TDC ($F[1,44] = 49.02$, $p < 0.001$, $\eta^2_p = 0.53$); a significant two-way interaction indicated that this deviation was more pronounced on certain tasks ($F[6, 264] = 4.51$, $p < 0.001$, $\eta^2_p = 0.09$). As shown in Figure 2, the follow-up analyses using *t*-tests revealed that children with unilateral CP had larger deviations than controls on all tasks and elicited very large differences between children with unilateral CP and controls, except the reach-forwards task.

CORRELATION BETWEEN KINEMATIC INDICES AND THE ASSISTING HAND ASSESSMENT SCORE

As shown in Figure 3, the upper limb global deviation (Global-APS) was highly predictive of bimanual performance (AHA; $r = -0.75$, $p < 0.001$). This global index and AHA score also depended on the functional level of the affected hand (MACS levels).

Table I presents the correlation coefficients between the various kinematic indices (Global-AVS or APS) and bimanual performance (AHA). Correlations between the AHA and the segmental deviation (Global-AVS) was high for wrist flexion, good for elbow flexion, and fair but significant for pronation and shoulder flexion.

Correlations between upper limb movement deviations and hand function were not the same for all the tasks (Table I). The correlation between AHA and APS was high for the reach-to-grasp tasks, high-to-moderate for the gross motor tasks, and fair-to-weak for the reach tasks.

DISCUSSION

KINEMATIC ANALYSIS BETWEEN CHILDREN WITH UNILATERAL CEREBRAL PALSY AND THE TYPICALLY DEVELOPING CHILDREN

Kinematic indices in children with unilateral CP and TDC Similar kinematic index values to those reported by Jaspers et al. were found¹¹, confirming that children with unilateral CP have significantly higher movement deviations of their impaired upper limb compared with the non-dominant limb of the TDC. Kinematic abnormalities were more pronounced on the distal segments (elbow flexion-extension, pronosupination and wrist flexion-extension). As can be seen in Figure S2 (online supporting information), the unilateral CP group tended to perform the unimanual tasks with more pronation, wrist flexion, ulnar deviation, and less elbow extension than the TDC group. These results are in line with previous 3DMA studies^{9,10}.

One explanation could be that the muscle weakness and the increased muscle tone are more prominent distally⁸, and have a higher influence on distal movements²¹. Moreover, the upper limb is a complex kinematic chain with a lot of degrees of freedom; we could imagine that small deviations in the proximal segments (i.e. trunk or shoulder) lead to large deviations in the more distal segments.

CORRELATION BETWEEN KINEMATIC INDICES OF THE AFFECTED UPPER LIMB AND BIMANUAL PERFORMANCE

The relationship between unimanual capacity and bimanual performance in children with unilateral CP interests the scientific community, as exemplified by a previous study⁶. Our research provides complementary information by establishing the correlation between the movement deviations during unimanual tasks and bimanual performance. A strongly negative correlation was found between bimanual performance (AHA) and kinematic abnormalities of the impaired upper limb (Global-APS). The higher the global movement deviation was, the lower the bimanual performance and the manual ability levels were. Furthermore, bimanual performance was strongly correlated to the segmental deviation (Global-AVS) of wrist and elbow flexions. Distal joint mobility seems to have a greater impact on the bimanual function than proximal joint mobility (shoulder and trunk). Indeed, the performance of the assisting hand is probably related to the child's ability to effectively position his or her forearm to stabilize an object in the impaired hand so that the non-impaired hand can use it. These findings support the fact that the corticospinal tract plays a disproportionate role in controlling distal function²². Deviations of the trunk and shoulder seem to act as compensatory mechanisms, which try to help the good positioning of the forearm and the hemiplegic hand. These results stress the importance of taking into account the distal joint movements in therapeutics.

Despite of a pronounced kinematic deviation for the pronosupination in children with unilateral CP compared to the TDC, a low correlation was shown with bimanual performance. In agreement with the clinical impression, a higher relationship could be expected for this degree of freedom. A possible explanation could be that the lack of active supination in children with unilateral CP is partly compensated by proximal joints such as shoulder and trunk⁹.

These results were reinforced by a recent study of Maillieux et al.²¹, which found similar values of correlation between the AHA score and the kinematic indices (AVS and APS) in reach-to-grasp vertically, hand-to-mouth and hand-to-head tasks. This cited work studied fewer tasks than in our work (three vs seven) and proposed results dedicated to each task separately. However, the functionality of the upper limb is not restricted to a single task but to multiple possibilities such as grasping, manipulating, reaching, and so on. Therefore, the scientific positioning of our study was to provide a global and realistic vision of impaired motor control of the upper limb, by calculating global kinematic indices on a set of seven complementary tasks.

In our study, the global movement deviation of the affected upper limb explains only 57% of the AHA variance. Two possible explanations can be mentioned. Firstly, gross motor and reach-to-grasp tasks were highly correlated to the AHA, whereas the reach tasks were, at best, only weakly correlated. A stronger correlation between the AHA and the Global-APS should be found by excluding these two reach tasks. Secondly, the impaired motor control in the affected upper limb is not the only parameter that influences the bimanual performance. Indeed, previous studies showed that AHA variance was also partially explained by grip strength⁸, spatial tactile deficiency⁷, stereognosis⁶, and synkinesis²³. Generally speaking, it suggests that tasks that require complex motor planning (e.g. to grasp and manipulate, with and without both hands) may be more relevant to assess the functionality of the impaired upper limb in 3DMA.

LIMITATIONS

Some critical reflections have to be made. First, the population studied was subject to inclusion and exclusion criteria from the current clinical research project aiming to assess the effect of mirror therapy in children with hemiplegia. Our population is nevertheless representative of the population of children suited to 3DMA, as protocol of Jaspers et al. has been validated in a unilateral CP group with similar inclusion criteria¹⁸. Secondly, scapular motion was not calculated because of difficulties in our calibration of the acromial cluster, which was not precise enough and was responsible for erroneous measurement. The International Society of Biomechanics' recommendations also have limitations as the markers are subject to displacement because of soft tissue artefacts. However, with regard to the findings of Maillieux et al. (21), which suggest that the scapular movement deviations were correlated neither to the bimanual performance (AHA) nor to the manual abilities (MACS levels), the impact of the scapular movements on the functionality of the upper limb can be questioned.

PERSPECTIVE

3DMA of the upper limb is complex because of the lack of cyclic movements, the variety of functions, and abundant degrees of freedom. Therefore, most 3DMA studies have analysed movement of the affected upper limb in standardized and unimanual situations. In the study by Maillieux et al.²¹, the kinematic indices (APS and distal AVS) were better correlated to the unimanual capacity than to the bimanual performance, highlighting that the 3DMA unimanual protocol assesses more the capacity of the affected hand (i.e. what the child can do with his/her affected arm on request). However, in everyday life, children with unilateral CP use their less affected hand to do unimanual tasks. More difficulties were observed in bimanual situations because of the limited motions of the "assisting" impaired hand and its repercussions on the less impaired limb⁴.

The AHA is presently considered as the best performance-based measure of bimanual activity in children with unilateral CP²⁴, but two limitations can be mentioned. Firstly, it is a subjective tool based on a video recording and scored by an operator. Secondly, it does not measure the combined functionality of the two hands (i.e. bimanual coordination) but rather the function of the affected hand in bimanual situations. AHA scores are thus highly correlated with unimanual measures of the affected hand function⁶. A more objective and quantitative description of the two upper limbs in a context of bimanual coordination can be obtained with 3DMA. Few studies have evaluated the kinematics of the hemiplegic limb in a bimanual situation^{25,26}, but none has explored the waveform of the movements by using kinematic indices. In futures studies, it would be important to develop a 3DMA bimanual protocol that assesses the interaction between both hands, with good reliability, validity, and sensitivity to change.

CONCLUSION

Bimanual performance of the children with unilateral CP is highly correlated to the movement deviations of the impaired upper limb, especially in distal joints. These findings stress the importance of taking into account the movements of wrist and elbow joints in therapeutics. In this context of 3DMA unimanual protocol, tasks that require more distal motion, such as reach-to-grasp or gross motor tasks, better reflect bimanual performance and appear as the key tasks to assess the functionality of the impaired upper limb. In a future study, it would be important to develop a 3DMA protocol in the context of bimanual tasks in order to assess the movement abnormalities of both impaired and non-impaired upper limbs in children with unilateral CP.

In a future study, it would be important to develop a 3DMA protocol in a context of bimanual tasks in order to assess the movement abnormalities of both the impaired and non-impaired upper limb in children with unilateral CP.

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SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section at the end of the article.

Figure S1: Position of the markers on the impaired limb of a child with unilateral cerebral palsy.

Figure S2: The average waveforms of the unilateral cerebral palsy group vs the typically developing children group.

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Table I : Correlation coefficients between kinematic indices (Global Arm Variable Score [AVS], Arm Profile Score [APS], Global-APS) and Assisting Hand Assessment score

Area	AVS score	<i>r</i>	<i>p</i> -value	APS score	<i>r</i>	<i>p</i> -value
Trunk	Flexion	-.10	.65	Reach forwards	-.37	.09
	Lateral flexion	-.53	.01	Reach sideways	-.49	.02
	Rotation	-.11	.62	Hand to mouth	-.75	<.001
Shoulder	Abduction	-.35	.10	Hand to head	-.73	<.001
	Flexion	-.48	.02	Hand to shoulder	-.58	.004
	Rotation	-.35	.11	Reach to grasp vertically	-.75	<.001
Elbow	Flexion	-.61	.002	Reach to grasp horizontally	-.75	<.001
	Pronation	-.47	.02	Average APS score	-.75	<.001
Wrist	Flexion	-.85	<.001			
	Deviation	-.20	.36			
Average AVS score		-.76	<.001			

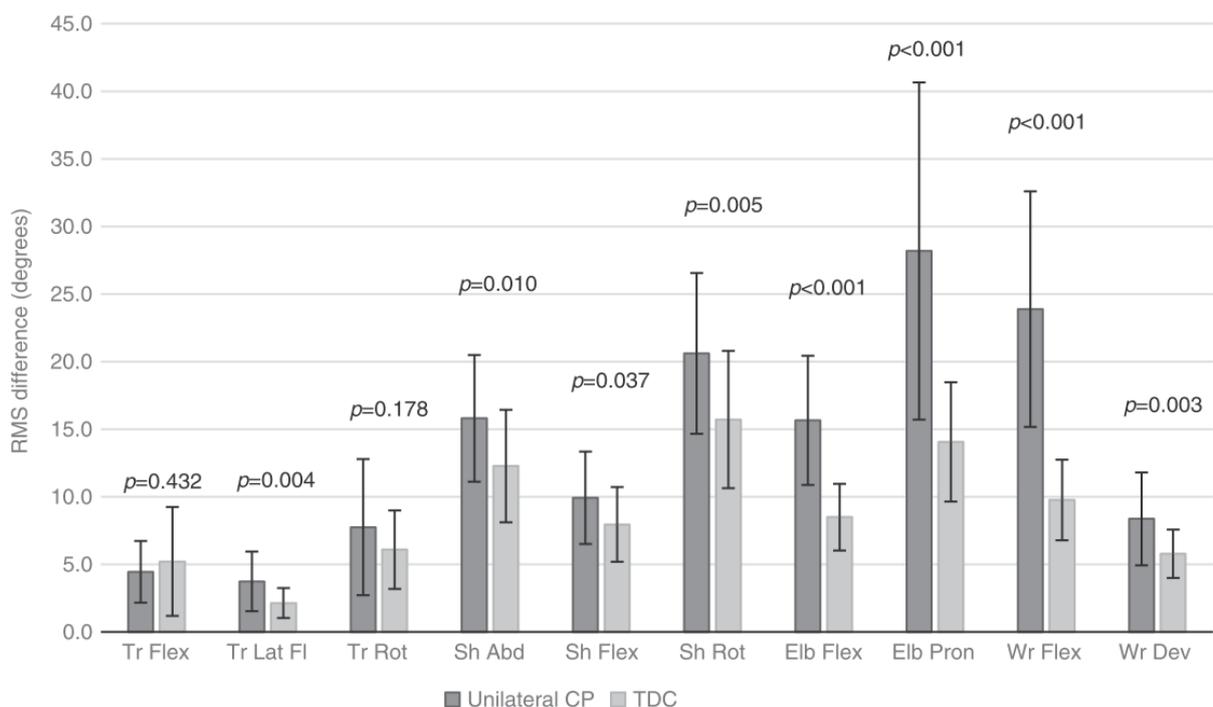


Figure 1: Global-Arm Variable Score (Global-AVS) in children with unilateral cerebral palsy (CP) and typically developing children (TDC). Error bars represent +/- 1 standard deviation around the mean. Significant differences between groups were determined using Welch 's t -test for unequal variances. RMS, root mean square; Tr, trunk; Flex, flexion; Lat FI, lateral flexion; Rot, rotation; Sh, shoulder; Abd, abduction; Elb, elbow; Pron, pronation; Wr, wrist, Dev, deviation.

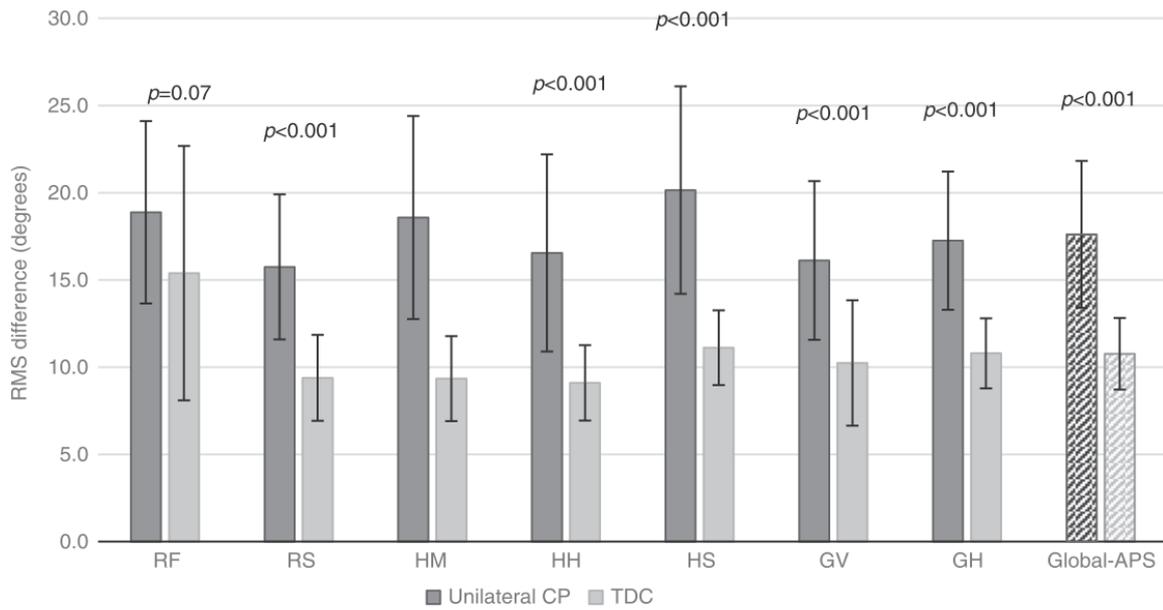


Figure 2: Arm Profile Score (APS) and Global-APS in children with unilateral cerebral palsy (CP) and typically developing children (TDC). Error bars represent +/-1 standard deviation around the mean. Significant differences between groups were determined using Welch 's t-test for unequal variances. RMS, root mean square; RF, reach forwards; RS, reach sideways; HM, hand to mouth; HH, hand to head; HS, hand to shoulder; GV, reach to grasp vertically; GH, reach to grasp horizontally.

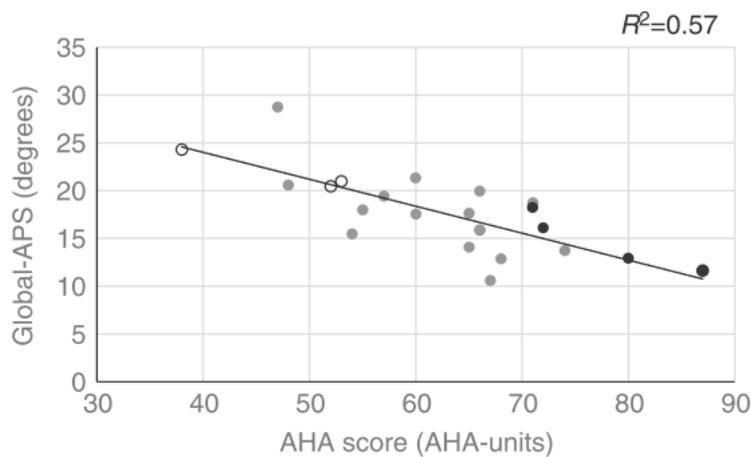


Figure 3: Relationship between the Global-Arm Profile Score (Global-APS) and the Assisting Hand Assessment (AHA) score. The line represents the slope of a linear regression model. Each patient is characterized by their Global-APS value and AHA score. Manual Ability Classification Scale (MACS) level is represented as black (MACS level I), grey (MACS level II), and white (MACS level III) circles.