

## PDF hosted at the Radboud Repository of the Radboud University Nijmegen

The following full text is a preprint version which may differ from the publisher's version.

For additional information about this publication click this link.

<http://hdl.handle.net/2066/167592>

Please be advised that this information was generated on 2020-11-30 and may be subject to change.

**Children with unilateral Cerebral Palsy show diminished implicit motor imagery with  
the affected hand**

*Marijtje L.A. Jongsma<sup>1</sup>, C. Marjolein Baas<sup>1</sup>, Anouk F.M. Sangen<sup>1</sup>, Pauline B.M. Aarts<sup>2</sup>, Rob H.J. van der  
Lubbe<sup>3,4</sup>, Ruud G.J. Meulenbroek<sup>5</sup> and Bert Steenbergen<sup>1,6</sup>*

<sup>1</sup>Behavioural Science Institute, Radboud University Nijmegen, P.O. Box 9104, 6500 HE Nijmegen, the  
Netherlands

<sup>2</sup>Department of Pediatric Rehabilitation, Sint Maartenskliniek, P.O. Box 9011, 6500 GM Nijmegen,  
The Netherlands

<sup>3</sup>Cognitive Psychology and Ergonomics, University of Twente, P.O. Box 217, 7500 AE, Enschede, the  
Netherlands

<sup>4</sup>Department of Cognitive Psychology, University of Finance and Management, Pawia 55, 01-030,  
Warsaw, Poland

<sup>5</sup>Donders Institute for Brain, Cognition, and Behaviour, Donders Centre for Cognition, Radboud  
University Nijmegen, P.O. Box 9104, 6500HE Nijmegen, the Netherlands

<sup>6</sup>Australian Catholic University, School of Psychology, Fitzroy Victoria 3065 Melbourne, Australia

**Corresponding author:**

Marijtje L.A. Jongsma  
Behavioural Science Institute  
Radboud University Nijmegen  
P.O.Box 9104  
6500HE Nijmegen  
the Netherlands  
tel: +31.(0)24.3616278

**This manuscript has been published in a revised version as:**

*Jongsma MLA, Baas CM, Sangen AMF, Aarts PBM, van der Lubbe RHJ, Meulenbroek RGJ,  
Steenbergen B. (2016) Children with unilateral cerebral palsy show diminished implicit motor  
imagery with the affected hand. *Developmental Medicine & Child Neurology*: 58(3); 277–284.  
DOI: 10.1111/dmcn.12819*

## **Abstract**

**Aims:** Motor imagery (MI) refers to the mental simulation of a motor action without producing an overt movement. Implicit MI can be regarded as a first-person kinesthetic perceptual judgment, and addresses the capacity to engage into the manipulation of one's body schema. In this study, we examined whether children with unilateral cerebral palsy are able to engage in implicit motor imagery.

**Methods:** A modified version of the hand laterality judgment task was employed. Erroneous responses, reaction times, and event-related potentials from the electroencephalograph were analyzed.

**Results:** In thirteen typically developing children, we observed the classic rotation direction effect. Specifically, when comparing outward rotated to inward rotated hand pictures, decreased accuracy and increased response times were observed. Event-related potentials analyses of the electroencephalogram revealed a more marked N1 and an enhanced rotation related negativity.

**Interpretation:** These findings suggest that an implicit motor imagery strategy was used to solve the task. However, in ten children with unilateral cerebral palsy, these effects were only observed when the less-affected hand was involved. This observation suggests that children with CP could benefit from visual training strategies.

### **What this paper adds: (5 bullets, 5-10 words per bullet)**

- Implicit motor imagery was studied in unilateral Cerebral Palsy
- Behavioral data and Event-related potentials quantified implicit motor imagery capacity
- Children with unilateral cerebral palsy engaged in implicit motor imagery
- However, implicit motor imagery was compromised for the affected hand.
- These findings might increase our understanding of body schema development

### **Short title:**

Implicit motor Imagery in Cerebral Palsy

---

Cerebral palsy (CP) describes a group of permanent disorders of movement and posture that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain<sup>1</sup>. In the current study we focused on children with unilateral CP with one hand being more affected than the other hand. Motor impairments associated with CP can be understood as a diminished ability of the brain to control complex motor programs<sup>2-5</sup>.

Motor imagery (MI) refers to the internal representation of an action without producing an overt body movement<sup>4-7</sup>. With respect to MI, a distinction between explicit and implicit MI can be made. During explicit MI a specific motor act is internally simulated whereas implicit MI refers to the ability to engage into the projection and manipulation of the body schema from a first person perspective<sup>6-8</sup>. An often used paradigm to test the implicit MI ability is Parsons' hand laterality judgment (HLJ) task<sup>6,9</sup>. In this forced choice task, participants judge the laterality of displayed hands as quickly as possible by determining whether a left or right hand is depicted. Typically, the reaction times (RTs) increase more for hand pictures that are rotated outward than inward. Parsons<sup>9</sup> proposed that prolonged RTs for outward rotated hands reflect the biomechanical constraints encountered when mentally rotating one's own hand to match the depicted hand stimulus. Thus, it is generally believed that participants engage in a kinesthetic mental rotation to solve the HLJ task<sup>6,7</sup>. This rotation effect has been repeatedly replicated since<sup>10-12</sup>.

In recent years, behavioral studies emerged that scrutinize the MI ability of individuals with CP<sup>13-15</sup>. It was concluded that although adolescents with CP are able to engage in mental rotation, the implicit MI capacity seemed to be compromised. Recently, Williams et al.,<sup>14,15</sup> found that children with unilateral CP were slower and less accurate on the HLJ task than a comparison group<sup>14,15</sup>.

Although implicit MI has been extensively researched by analyzing overt behavioral measures like RTs and response accuracy, these data only reflect the outcome of combined cognitive and response processes, rather than the isolated process itself. Extracting event-related potentials (ERPs) from the ongoing electroencephalogram (EEG) however provides an excellent means to directly study the neural responses associated with implicit MI<sup>11,12</sup>. ERP components are typically divided into two types, based on their respective latencies. Components with latencies of up to 100 ms are referred to as exogenous components<sup>16</sup> whereas the endogenous components (>100 ms after stimulus onset) are assumed to be determined by cognitive aspects of information processing<sup>16</sup>. In the current study we focused on these endogenous components. Previous ERP studies have shown that mental rotation is accompanied by a negative-going amplitude modulation of the late-latency ERP components<sup>10-12</sup>. This modulation has been referred to as rotation related negativity (RRN) and has been observed in several mental rotation studies<sup>10,11</sup>. Interestingly, RRN is more pronounced for outward rotated stimuli than for inward rotated stimuli<sup>11,12</sup>. These findings suggest that RRN, similar to RT, is also modulated by biomechanical constraints<sup>11,12</sup>.

Only a few studies have investigated implicit MI by means of EEG measurements in patient groups. For example, Van Elk et al.,<sup>17</sup> recorded the EEG of young adults with unilateral CP while they performed the HLJ task. Results revealed a reduced RRN over parietal areas and prolonged RTs for the CP group compared to controls. However, stroke patients with acquired hemiplegia seem to preserve their implicit MI ability for both their paralyzed and non-affected hand<sup>18,20</sup>.

Unlike stroke patients, adolescents and children with unilateral CP lack a typical early development of the body schema<sup>8</sup>. Moreover, their internal body representation might be less accurate with respect to the affected side of the body than of the less-affected side. Because implicit MI relies on the ability to engage into the projection and manipulation of the body schema from a first person perspective, we hypothesize that in unilateral CP, implicit MI is especially compromised when the affected hand is involved, but less so when the less-affected hand is involved<sup>7</sup>. To the best of our knowledge, previous research has never compared the implicit MI capacity of the affected and less-affected hand separately in children with unilateral CP.

In the present study, our main aim was to determine if implicit MI capacity is (partly) determined by the motor capacity of the involved hand. We did this by recording ERPs that were elicited in response to the Parsons' HJL task. Response accuracy and response speed were also measured.

## **Materials and Methods**

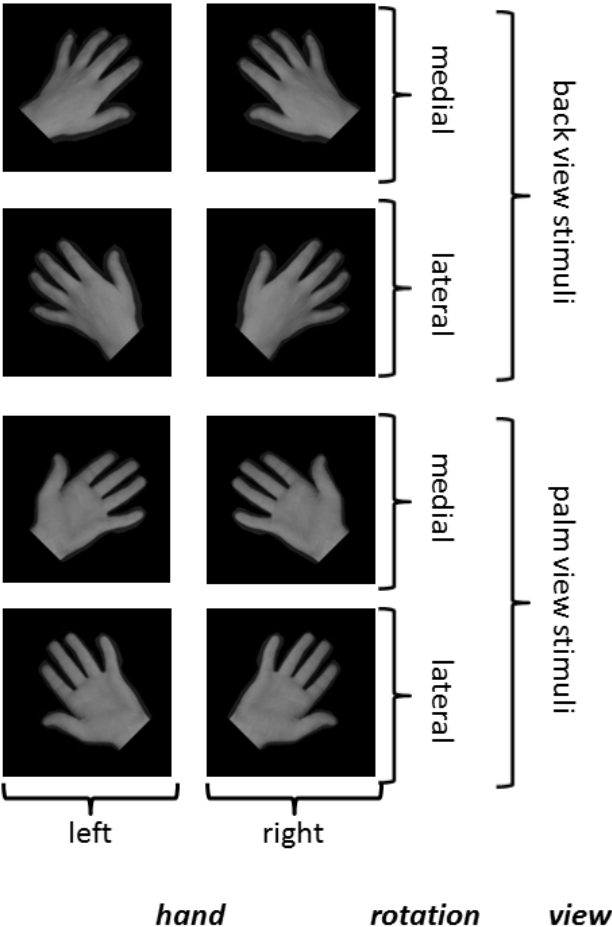
Informed consent was obtained prior to the start of the experiment and the followed procedures were approved by the local ethics committee (nr. ECG30062011). Ten children with unilateral CP (mean age 10 years, 7 months, SD 2 years, 5 months; 5 male; 4 left hand affected, all with IQ scores >70) participated in the study (CP group) and thirteen typically developing children (mean age 10 years, 7 months, SD 1 years, 2 months; 7 male; all right handed) participated in the experiment. There was no evidence that groups differed in the proportion of each gender or the mean age. ~~Groups did not differ with respect to gender and age.~~

An adapted version of Parson's HLJ task was employed<sup>11</sup> to study the implicit MI ability. Participants' hands were covered with a cloth in order to prevent a visual matching strategy. Participants' hands were positioned over a large response button (diameter: 9.5 cm; height: 5.5 cm) to capture both the RTs and laterality decisions. Visual stimuli, presented on the screen, consisted of photos of a child's left or right hand. The hand was seen from either a palm view or a back view perspective, and was rotated by 60° in either an inward or outward direction. This resulted in eight different stimuli (see figure 1a). Participants had to generate a response upon presentation of a visual response screen, which was displayed after a fixed waiting interval of 1700 ms (delayed response task). By introducing a delayed response we expected MI to be less distorted by cognitive processes related

to response execution and concurrent motor artifacts. Participants were asked to judge the laterality of the displayed hands as quickly and accurately as possible after the response screen by pressing either the left or right response button. Participants were only instructed to judge the hand laterality and were not instructed to use motor imagery to solve the task. In total 96 trials were presented. The experiment lasted about 15 minutes. An example of the setup of a trial is shown in figure 1b.

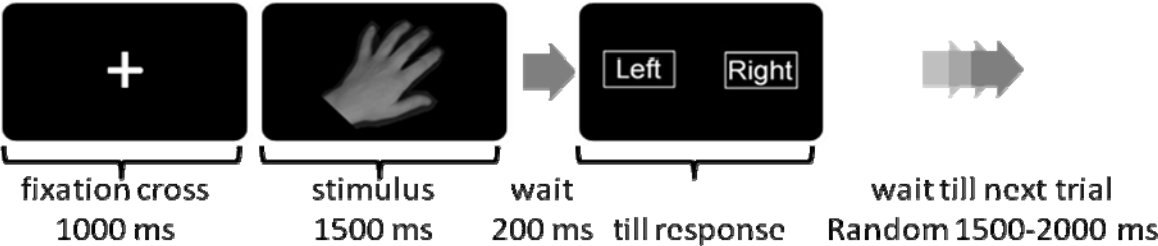
**Figure 1a: Stimulus material**

*The eight different hand stimuli that were randomly presented during the experiment.*



**Figure 1b: Task.**

*Graphical representation of a trial in the adapted hand laterality judgment task.*



EEG and electrooculographic (EOG) signals were recorded with a 32-channel actiCap system (Brain Products GmbH, Munich, Germany). Electrodes were located on positions according to the international 10-20 system. Measured activity was referenced to linked mastoids<sup>10,19</sup>. A ground electrode was located at the AFz electrode position. Electrode impedance was kept below 5 k $\Omega$ . Eye movements were recorded by electrodes placed below the right eye and at the outer canthus of the right eye. The signal was online digitized at 1000 Hz, with a high-pass and low-pass filter set at 0.1 Hz and 100 Hz, respectively. For children with CP that were right-hand affected (n=4) the electrode positions were inverted (i.e., P3 was redefined as P4, etc.). The EEG was corrected for EOG artifacts by employing the Gratton and Coles algorithm. High-pass and low-pass filters of 0.53 Hz and 40 Hz were subsequently applied. EEG data on trials with incorrect responses or trials contaminated with artifacts exceeding an amplitude of 150  $\mu$ V detected by automatic segment selection provided by Brain Vision Analyser were also excluded (for the TD and CP group 4% and 12% of the total amount of trials, respectively). A 250 ms interval was used for baseline correction. Next, averages were computed per stimulus type. Grand average ERPs were additionally computed for each group, which are displayed in figure 2a-b.

Accuracy was determined by analyzing the percentage of errors with a repeated measures ANOVA with the variables rotation (inward or outward rotation) and view (palm view or back view) as within subject factors, and group (CP or TD) as between subjects factor.

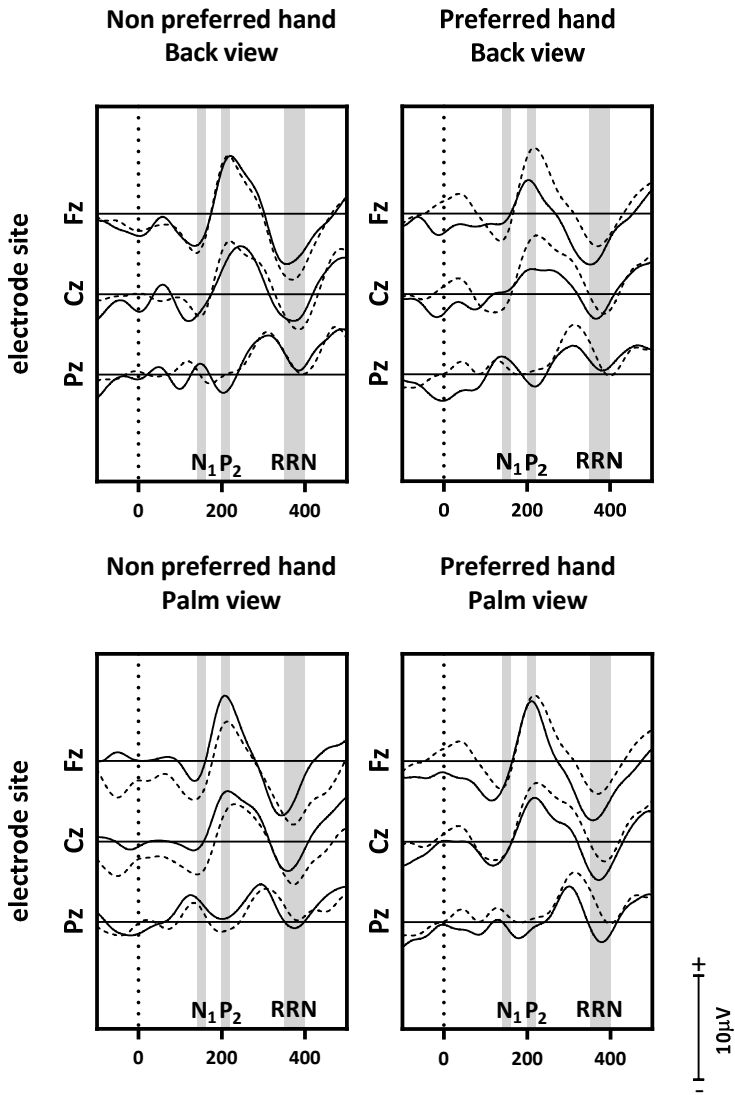
For average error percentage analysis, nonparametric Wilcoxon signed-rank tests and Mann-Whitney tests were conducted since error data did not meet parametric assumptions. Reaction times were also analyzed with a repeated measures ANOVA with the variables hand rotation and view as within subject factors, and group as between subjects factor. Whenever interaction effects were observed, appropriate post-hoc tests were performed.

After visual inspection of the grand average ERPs the N1, P2, and RRN components could be identified. ERP amplitudes were determined as the average value within a fixed latency window (N1 140-160 ms; P2 200-220 ms; RRN amplitude 350-400 ms; Figure 2).

**Figure 2a: Grand average event-related potentials (ERPs) of the group of typically developing (TD) children**

ERPs are depicted for the midline electrodes Fz, Cz, Pz on the y-axis. Medially rotated hand stimuli are depicted with a dotted line whereas laterally rotated stimuli are depicted with a solid line. The x-axis shows the time related stimulus onset (in ms). Grey bars mark the N1, P2 and RRN latency windows. The upper panels show the ERPs for the back view stimuli with on the left ERPs to stimuli depicting the non preferred hand and on the right ERPs to stimuli depicting the preferred hand. The lower panels show the ERPs for the palm view stimuli with on the left ERPs to stimuli depicting the non preferred hand and on the right ERPs to stimuli depicting the preferred hand.

**Typically developing children**

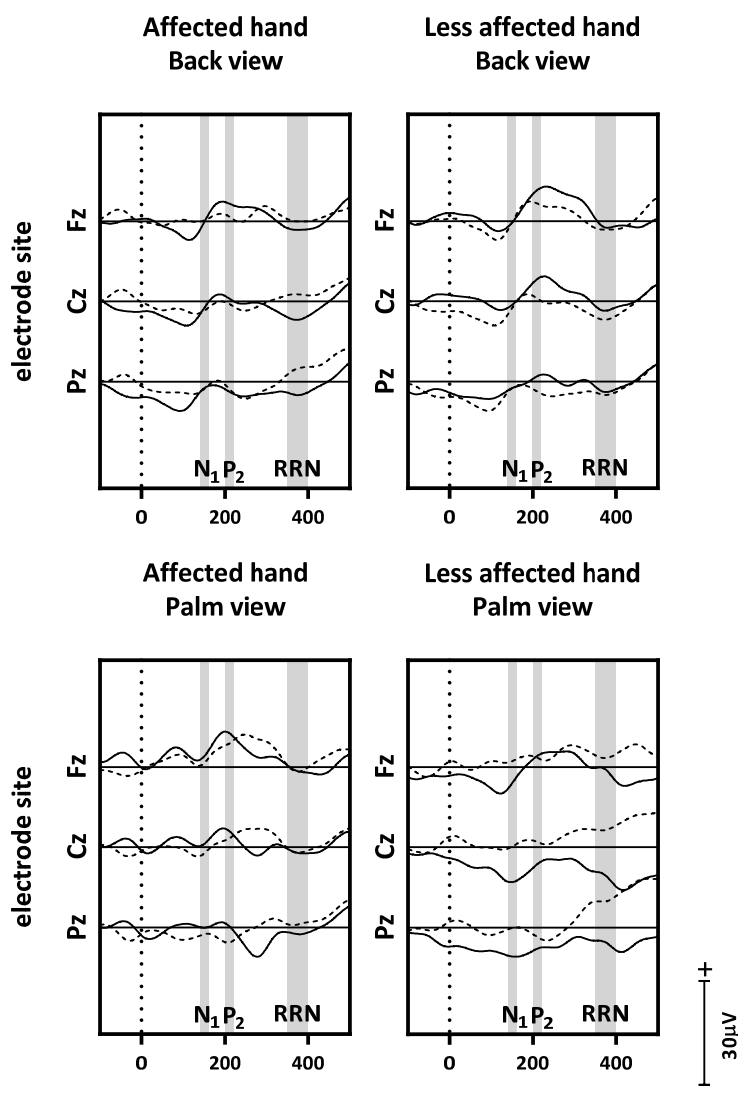




**Figure 2b: Grand average event-related potentials (ERPs) of the group of children with unilateral Cerebral Palsy (CP)**

ERPs are depicted for the midline electrodes Fz, Cz, Pz on the y-axis. Medially rotated hand stimuli are depicted with a dotted line whereas laterally rotated stimuli are depicted with a solid line. The x-axes show the time related stimulus onset (in ms). Grey bars mark the N1, P2 and RRN latency windows. The upper panels show the ERPs for the back view stimuli with on the left ERPs to stimuli depicting the affected hand and on the right ERPs to stimuli depicting the less-affected hand. The lower panels show the ERPs for the palm view stimuli with on the left ERPs to stimuli depicting the affected hand and on the right ERPs to stimuli depicting the less-affected hand.

**Children with unilateral CP**



The N1 and P2 appeared to be maximal over the frontal region and the data from F3/Fz/F4 were further analyzed. The RRN effect seemed maximal over the parietal region and the data from P3/Pz/P4

were further analyzed. ERP component amplitudes were analyzed using a repeated measures ANOVA with the variables hand (left (non-preferred) versus right (preferred) hand), rotation (inward versus outward rotation), view (palm view versus back view), and electrode position with respect to the presented hands stimulus (ipsilateral: F/P3 for left and F/P4 for right hand stimuli; central: F/Pz; contralateral: F/P3 for left and F/P4 for right hand stimuli). Group served as a between subjects factor.

## Results

With respect to the error analyses, three participants with unilateral CP were excluded from further analysis because their performance did not exceed chance levels. Error analyses of the remaining participants revealed a main group effect and a main view effect, which reflected increased error rates for the CP group, and increased error rates for palm view stimuli. In addition, we observed a group\*hand interaction effect and a group\*view interaction effect. Results were further analyzed with nonparametric tests. The main effects of view and group remained significant, showing that CP children made more errors than typically developing children. Within the group of typically developing children no further effects were observed. Participants with CP made more errors to palm view stimuli ( $p < .05$ ) and to hand stimuli corresponding to the less-affected hand ( $p < .05$ ). In addition, they showed a trend towards a rotation\*hand interaction effect, indicating more errors for laterally rotated hand stimuli than for medially rotated hand stimuli, but only when stimuli depicted the less-affected hand ( $p < .1$ ). See table 1a for F(df), p, and  $\eta^2$  values. See figure 3 for the percentages of errors.

**Table 1a: Statistics error analyses.**

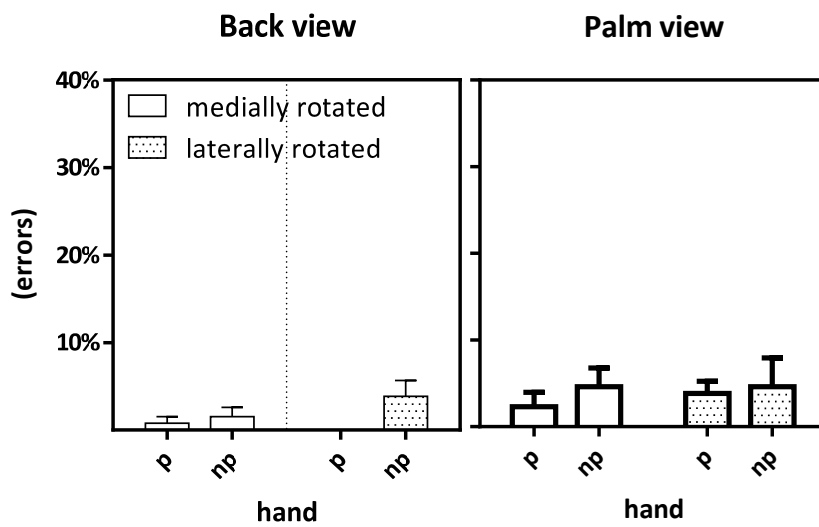
<b>Errors</b>	<b>F (df)</b>	<b>p</b>	<b><math>\eta^2</math></b>
Group	(1,18) =8.92	.01	.33
View	(1,18)=14.28	.001	.44
Group*Hand	(1,18)=5.63	.029	.24
Group*View	(1,18)=4.73	.043	.21
<b>Non parametric</b>		<b>Exact p</b>	<b>z</b>
Group	U=11.0	.004	-2.77
View		.005	-2.68

**Figure 3: Error analyses**

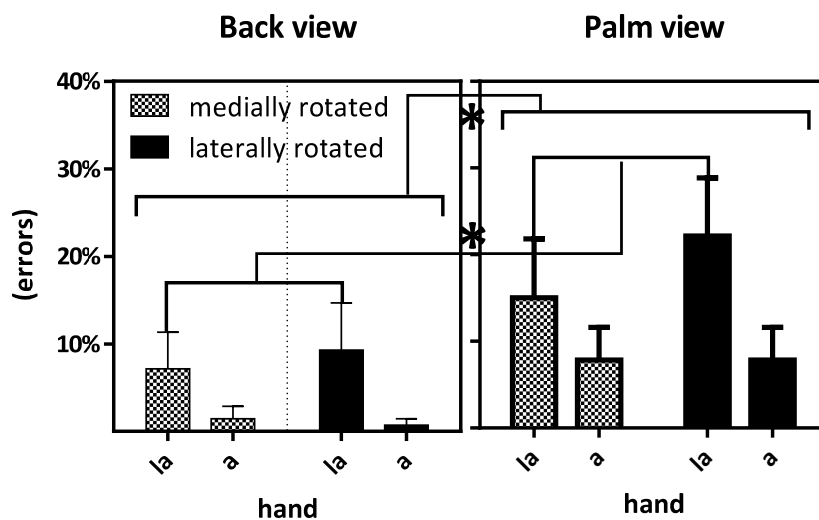
The upper panels show the bar graphs of the percentage of errors for the TD group depicted by light shaded bars. Medially rotated stimuli are depicted with white bars whereas laterally rotated stimuli are depicted by dotted bars. Back view stimuli are depicted with light lined bars whereas palm view stimuli are depicted by bold lined bars. The percentages are depicted for the preferred (p) and non preferred (np) hand. Asterisks mark the significances ( $P < .05$ ).

The lower panels show the bar graphs of the percentage of errors for the CP group depicted by dark shaded bars. Medially rotated stimuli are depicted with patterned bars whereas laterally rotated stimuli are depicted by black bars. Back view stimuli are depicted with light lined bars whereas palm view stimuli are depicted by bold lined bars. The percentages are depicted for the less-affected (la) and affected (a) hand. Asterisks mark the significances ( $P < .05$ ).

### typically developing children



### unilateral Cerebral Palsy



For response times, several previously reported RT effects were present despite the 1700 ms delayed response interval. RTs showed a main effect of view and a view\*rotation interaction. Post hoc tests per view indicated that RTs for outward rotated stimuli were slower than RTs for inward rotated stimuli, but only for palm view stimuli ( $p < .05$ ) and only in the TD group. See table 1b for  $F(df)$ ,  $p$ , and  $\eta^2$  values. See figure 4 for the percentages of errors.

**Table 1b: Statistics Response times.**

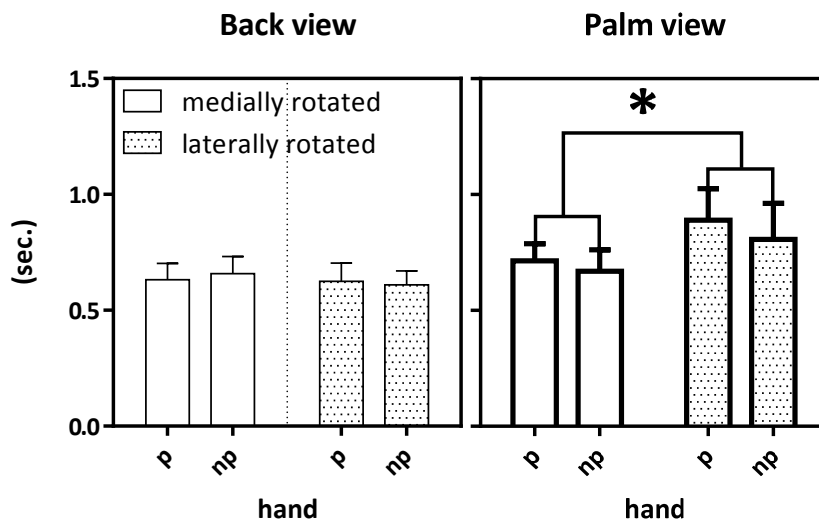
<b>RTs</b>	<b>F (df)</b>	<b>p</b>	<b><math>\eta^2</math></b>
View	(1,18)=13.57	.002	.43
View*Rotation	(1,18)=6.39	.021	.26

**Figure 4: Response time analyses**

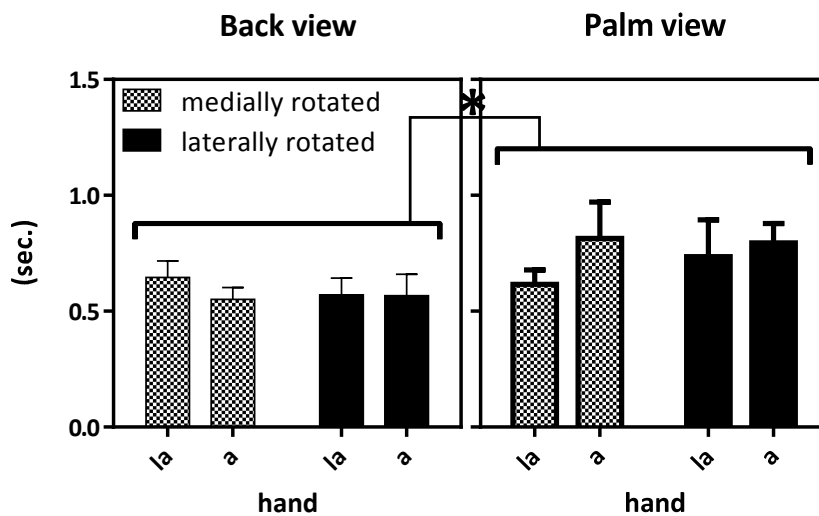
The upper panels show the bar graphs of the reaction times (RTs) for the TD group depicted by light shaded bars. Medially rotated stimuli are depicted with white bars whereas laterally rotated stimuli are depicted by dotted bars. Back view stimuli are depicted with light lined bars whereas palm view stimuli are depicted by bold lined bars. The percentages are depicted for the preferred (p) and non preferred (np) hand. Asterisks mark the significances ( $P < .05$ ).

The lower panels show the bar graphs of the reaction times (RTs) for the CP group depicted by dark shaded bars. Medially rotated stimuli are depicted with patterned bars whereas laterally rotated stimuli are depicted by black bars. Back view stimuli are depicted with light lined bars whereas palm view stimuli are depicted by bold lined bars. The percentages are depicted for the less-affected (la) and affected (a) hand. Asterisks mark the significances ( $P < .05$ ).

### typically developing children



### unilateral Cerebral Palsy



With respect the ERPs, the N1, P2 and RRN were further analyzed. For the N1 component, a hand\*view interaction and a group\*hand\*rotation electrode interaction were observed. Post-hoc analyses per group and view revealed a rotation\*electrode effect for palm view stimuli ( $p < .05$ ) within the TD. That is, the N1 was increased for outward rotated hand stimuli, especially over the contralateral electrode. In the CP group, no effects on the N1 component were observed.

A main electrode effect was observed for the P2 component with maximal values over Fz. In addition, several interactions with group were found: a group\*electrode interaction, a group\*hand\*view interaction a group\*hand\*view\*rotation interaction. However, post-hoc analyses only showed some minor electrode effects for back view stimuli in both groups suggesting higher amplitudes over the midline electrode.

RRN demonstrated a main rotation effect which reflected an increased RRN for outward rotated stimuli compared to inward rotated stimuli. In addition, several interaction effects with rotation were found: a hand\*rotation interaction, a group\*hand\*rotation interaction, and a group\*hand\*view\*rotation interaction. For electrode a group\*electrode interaction and a hand\*electrode interaction were observed. Post-hoc analyses per group revealed a main effect of rotation direction ( $p < .05$ ) within the TD group, confirming more pronounced RRNs for outward rotated hand stimuli than for inward rotated hand stimuli. In the CP group, a hand\*rotation ( $p < .05$ ) and a hand\*electrode interaction was observed ( $p < .05$ ). Post-hoc comparisons indicated a clear rotation effect for the less-affected, but not for the affected hand, again with more pronounced RRNs for outward rotated hand stimuli than for inward rotated hand stimuli. See table 1c for F(df), p, and  $\eta^2$  values. See table 2 for the ERP component amplitudes (means and SDs).

**Table 1c: Statistics ERP components.**

N1	F (df)	p	$\eta^2$
Hand*View	(1,18) =4.44	.049	.20
Group*Hand*Rotation*Electrode	(2,17)=4.88	.029	.37
Electrode	(2,17) =22.20	<.001	.72
Group*Electrode	(2,17)=3.99	.038	.32
Group*Hand*View	(1,18)=6.77	.018	.27
Group*Hand*View*Rotation	(1,18)=5.40	.032	.23
Rotation	(1,18)=6.43	.021	.26
Hand*Rotation	(1,18)=11.01	.004	.38
Group*Hand*Rotation	(1,18)=5.93	.026	.25
Group*Hand*View*Rotation	(1,18)=5.52	.030	.24

**Table 2a: ERP component amplitudes: means (SDs). Group of typically developing children.**

<b>N1 back view</b>	<b>Left outward</b>	<b>Left inward</b>	<b>Right inward</b>	<b>Right outward</b>
F <sub>contralateral</sub>	-1.6 (3.81)	-3.4 (3.96)	-4.4 (5.83)	-2.5 (3.40)
F <sub>midline</sub>	-3.6 (3.60)	-4.9 (5.54)	-3.6 (4.45)	-1.1 (3.85)
F <sub>ipsilateral</sub>	-3.1 (2.19)	-3.9 (5.60)	-2.8 (4.21)	-1.0 (4.53)
<b>N1 palm view</b>	<b>Left outward</b>	<b>Left inward</b>	<b>Right inward</b>	<b>Right outward</b>
F <sub>contralateral</sub>	-2.0 (3.44)	-6.1 (4.25)	-1.7 (2.84)	-3.3 (4.15)
F <sub>midline</sub>	-3.6 (4.37)	-3.1 (3.68)	-6.2 (3.98)	-1.8 (3.91)
F <sub>ipsilateral</sub>	-3.5 (4.68)	-4.0 (3.26)	-6.0 (3.92)	-1.4 (3.21)

<b>P2 back view</b>	<b>Left outward</b>	<b>Left inward</b>	<b>Right inward</b>	<b>Right outward</b>
F <sub>contralateral</sub>	5.5 (4.37)	6.4 (6.72)	5.6 (3.77)	4.0 (6.10)
F <sub>midline</sub>	6.6 (4.66)	6.9 (6.71)	7.2 (4.92)	5.0 (5.78)
F <sub>ipsilateral</sub>	5.1 (3.35)	6.5 (6.50)	6.1 (4.60)	3.8 (6.48)
<b>P2 palm view</b>	<b>Left outward</b>	<b>Left inward</b>	<b>Right inward</b>	<b>Right outward</b>
F <sub>contralateral</sub>	7.8 (4.23)	5.8 (4.88)	5.2 (8.65)	7.7 (4.08)
F <sub>midline</sub>	8.3 (3.66)	6.3 (5.64)	6.5 (7.73)	8.5 (4.65)
F <sub>ipsilateral</sub>	6.8 (3.03)	4.8 (5.22)	5.4 (7.18)	7.1 (3.20)

<b>RNN back view</b>	<b>Left outward</b>	<b>Left inward</b>	<b>Right inward</b>	<b>Right outward</b>
F <sub>contralateral</sub>	1.1 (6.08)	3.1 (4.38)	1.6 (4.07)	-1.3 (4.90)
F <sub>midline</sub>	-0.7 (4.69)	-0.1 (3.51)	0.3 (5.13)	1.4 (6.03)
F <sub>ipsilateral</sub>	-2.4 (8.23)	-1.6 (4.55)	0.5 (3.85)	-2.0 (6.38)
<b>RNN palm view</b>	<b>Left outward</b>	<b>Left inward</b>	<b>Right inward</b>	<b>Right outward</b>
F <sub>contralateral</sub>	-0.7 (4.30)	-0.1 (6.05)	-1.0 (5.46)	-2.00 (5.21)
F <sub>midline</sub>	-2.3 (4.85)	0.2 (4.27)	1.5 (4.27)	-0.6 (5.31)
F <sub>ipsilateral</sub>	-2.7 (3.65)	-2.0 (3.77)	-1.5 (5.47)	-2.4 (4.99)

**Table 2b: ERP component amplitudes: means (SDs). Group of children with Cerebral Palsy.**

<b>N1 back view</b>	<b>Left outward</b>	<b>Left inward</b>	<b>Right inward</b>	<b>Right outward</b>
F <sub>contralateral</sub>	-0.8 (7.98)	-4.5 (4.36)	-1.9 (10.09)	-3.8 (7.96)
F <sub>midline</sub>	-2.4 (4.02)	-0.9 (7.25)	-1.4 (7.44)	-0.8 (6.39)
F <sub>ipsilateral</sub>	-3.0 (5.33)	-3.3 (3.47)	-1.8 (10.11)	-2.9 (6.32)
<b>N1 palm view</b>	<b>Left outward</b>	<b>Left inward</b>	<b>Right inward</b>	<b>Right outward</b>
F <sub>contralateral</sub>	-11.9 (14.85)	0.3 (11.70)	-6.9 (9.13)	-2.9 (6.54)
F <sub>midline</sub>	-1.0 (8.78)	-1.1 (11.43)	2.7 (14.02)	-7.8 (8.50)
F <sub>ipsilateral</sub>	1.6 (12.08)	0.1 (9.52)	-0.5 (10.24)	-7.5 (7.75)

<b>P2 back view</b>	<b>Left outward</b>	<b>Left inward</b>	<b>Right inward</b>	<b>Right outward</b>
F <sub>contralateral</sub>	4.6 (4.02)	3.5 (11.13)	1.7 (7.40)	6.5 (7.90)
F <sub>midline</sub>	7.2 (4.45)	2.6 (11.73)	4.5 (7.14)	10.1 (9.01)
F <sub>ipsilateral</sub>	4.1 (6.63)	0.3 (9.70)	2.0 (6.31)	6.3 (8.67)
<b>P2 palm view</b>	<b>Left outward</b>	<b>Left inward</b>	<b>Right inward</b>	<b>Right outward</b>
F <sub>contralateral</sub>	9.7 (6.57)	2.3 (5.89)	2.3 (4.63)	-5.1 (17.37)
F <sub>midline</sub>	12.5 (9.41)	7.3 (11.81)	6.3 (5.55)	2.7 (5.22)
F <sub>ipsilateral</sub>	9.3 (8.75)	4.2 (10.19)	3.0 (1.87)	2.5 (4.43)

<b>RNN back view</b>	<b>Left outward</b>	<b>Left inward</b>	<b>Right inward</b>	<b>Right outward</b>
F <sub>contralateral</sub>	-2.8 (11.32)	1.4 (8.36)	5.2 (5.45)	2.9 (8.06)
F <sub>midline</sub>	1.7 (13.35)	5.3 (5.61)	2.1 (4.40)	-2.6 (13.66)
F <sub>ipsilateral</sub>	-2.3 (10.46)	1.5 (5.13)	2.0 (9.95)	0.9 (9.94)
<b>RNN palm view</b>	<b>Left outward</b>	<b>Left inward</b>	<b>Right inward</b>	<b>Right outward</b>
F <sub>contralateral</sub>	-5.1 (16.62)	8.7 (8.26)	13.0 (8.22)	-1.5 (14.25)
F <sub>midline</sub>	2.6 (8.61)	5.6 (17.36)	-0.4 (17.68)	-2.8 (12.02)
F <sub>ipsilateral</sub>	-1.3 (11.31)	-0.8 (17.04)	8.8 (10.86)	-2.7 (20.90)

## Discussion

In the present study, we investigated the implicit MI ability of children with unilateral CP compared to typically developing children. To study implicit MI capacity, an adapted version of the HLJ task<sup>11</sup> was used to capture ERPs together with overt measures of speed and accuracy. Our main aim was to determine if children with unilateral CP are capable to engage in an implicit MI task, and if so, if implicit MI capacity would depend on whether the affected hand or the unaffected hand was involved in this task. This issue has, to our knowledge, not been addressed before.

We hypothesized that the group of typically developing children would demonstrate an increase in errors and/or response times together with a more marked ERP components in reaction to outward rotated hand stimuli as compared to inward rotated hand stimuli<sup>10-12</sup>. Indeed, typically developing children showed these expected effects with respect to both the response times, the N1 component, and RRN, suggesting the use of an implicit MI strategy to solve this task. However, in the group of children with unilateral CP these effects were only observed when depicted stimuli were associated with the less-affected hand. Thus, children with unilateral CP seem capable in engaging in implicit MI, but less so when the affected hand is subject of the imagery task.

Children with unilateral CP appeared to be less accurate than typically developing children as evidenced by inflated error rates. Of note, within the current study, all typically developing children were able to perform the HLJ task above chance level. Within the group of children with unilateral CP, however, three participants performed at chance level, which we therefore removed from further analyses. For the remaining participants, the group with unilateral CP made significantly more errors than the group with typically developing children, and especially with respect to palm view stimuli. This accords with previous studies that reported a diminished implicit MI capacity in children and adolescents with CP<sup>5,7,13</sup>. Interestingly, children with unilateral CP made fewer errors when the presented hand picture corresponded to their affected hand. A similar effect has been described in stroke patients<sup>6</sup> and is known as an hemiplegic advantage<sup>18</sup>. Such an advantage might arise when a different strategy is adapted<sup>6</sup>. With implicit MI tasks, like the HLJ, participants may apply alternative strategies to reach a solution, for example, visual imagery or a 3<sup>rd</sup> person MI perspective approach may be used instead<sup>6</sup>. It has been proposed that when alternative strategies are used, task performance should be less-affected than when a 1<sup>st</sup> person kinematic approach would be adapted<sup>6</sup>. This is in line with our observation that children with CP seemed to be less erroneous when the affected limb was involved.

Although we employed a delayed response task, previously reported RT effects could still be observed. No group differences on RTs were observed suggesting that RTs were predominantly



determined by the difficulty of the task instead of general motor speed, in which case the children with unilateral CP should have displayed prolonged RTs. For both groups, longer RTs for palm view stimuli than for back view stimuli were observed. Due to the diminished visual familiarity with viewing the own hands from a palm view perspective, it has been proposed that participants are more likely to engage in MI when palm view stimuli are presented<sup>11,12</sup>.

As expected, with respect to palm view stimuli, typically developing children revealed prolonged RTs for outward compared to inward rotated stimuli. This commonly reported observation has previously been explained in terms of biomechanical constraints<sup>9-12</sup>. In the group of children with unilateral CP, this RT effect was only observed when stimuli depicted the less-affected hand, but not when the affected hand was depicted, suggesting a diminished MI capacity when the affected hand was involved.

With respect to the ERPs elicited by the hand pictures, a N1, a P2, and the classically reported RRN between 350-400 ms after stimulus presentation could be observed. Unexpectedly, but in line with the RT results, typically developing children had a more marked N1 to outward rotated stimuli compared to inward rotated stimuli for palm view stimuli. This may reflect an increased spatial attention and possibly an early activation of the contralateral located pre-motor areas<sup>16</sup>. In addition, typically developing children had a more marked RRN to outward rotated stimuli compared to inward rotated stimuli. This is in line with previous ERP research applying the HLJ task<sup>10-12</sup> and suggests the use of a 1<sup>st</sup> person kinematic approach to solve this task<sup>6</sup>.

Importantly, within the group of children with unilateral CP, no rotation effect on the ERP N1 was observed. In addition, the RRN effect for outward compared to inward rotated hand stimuli was only observed for stimuli depicting the less-affected hand but not for stimuli depicting the affected hand. Together with the RT results, these findings suggest that children with unilateral CP are capable to use a 1<sup>st</sup> person kinematic approach to solve the HLJ task when the less-affected hand is involved, but not when the affected hand is involved. Previous studies have suggested that implicit MI depends (in part) on the imagers' body scheme<sup>7,8</sup>. For example, it has been reported that hemiplegic post-stroke participants rely on visual strategies to compensate deficient access to their body schema<sup>7,8</sup>. Although previous research has reported a diminished MI capacity in general in both children and adolescents with CP and children with DCD<sup>22</sup>, the current study is the first study, to our knowledge, that reveals a hand specific decrease in MI capacity. Therefore, MI might be even more intertwined to internal body schema than previously assumed.

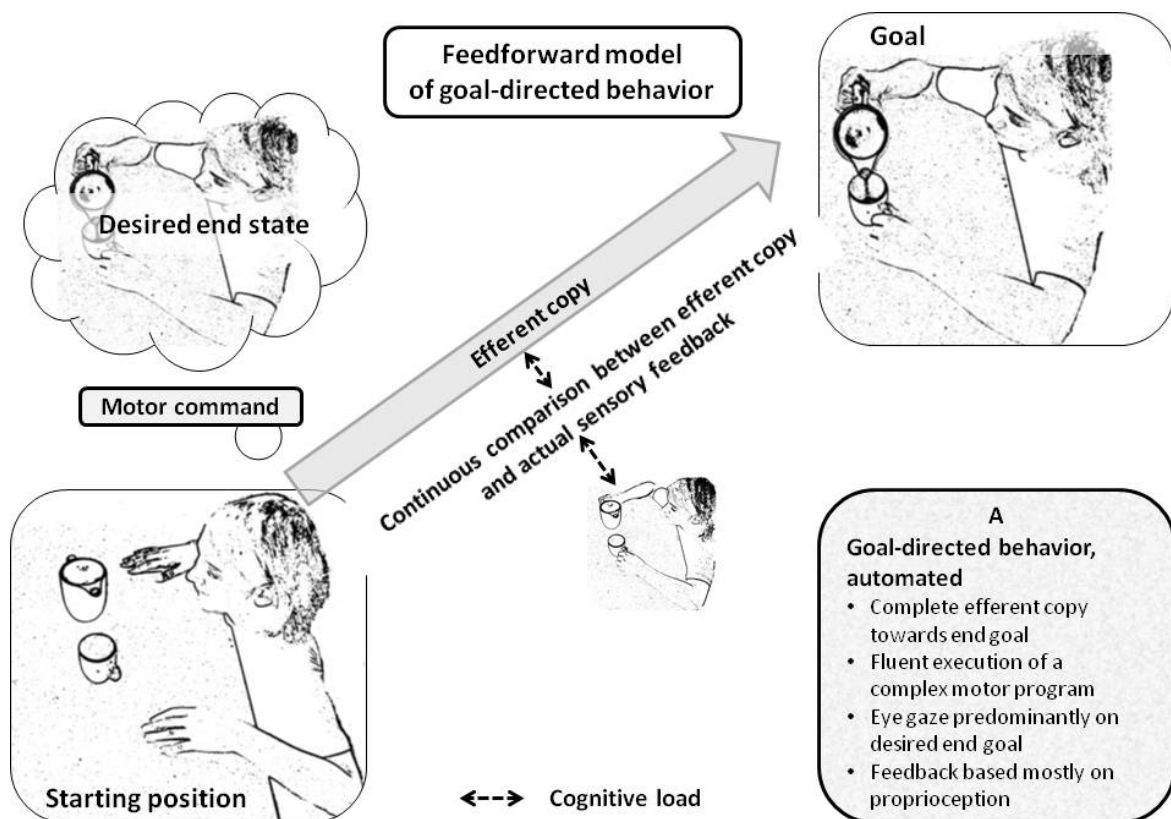
Finally, it has been reported that children and adolescents with CP devote a lot of visual attention to their affected limb<sup>23</sup>. For example, Steenbergen et al.,<sup>23</sup> reported that during bimanual actions visual attention seemed to be drawn to the affected side of the body in participants with unilateral CP. Others have demonstrated that for the on-line representation of the body schema both

proprioception and visual information are integrated<sup>7</sup>. By covering the hands, as is customary during the HLJ task, our group of children with CP could not fall back on a visual control approach when solving the HLJ task and are proposed to have reduced proprioceptive input from the affected hand resulting in diminished performance when the affected hand was involved.

See also the additional figures 5a and 5b below (not included in the final article).

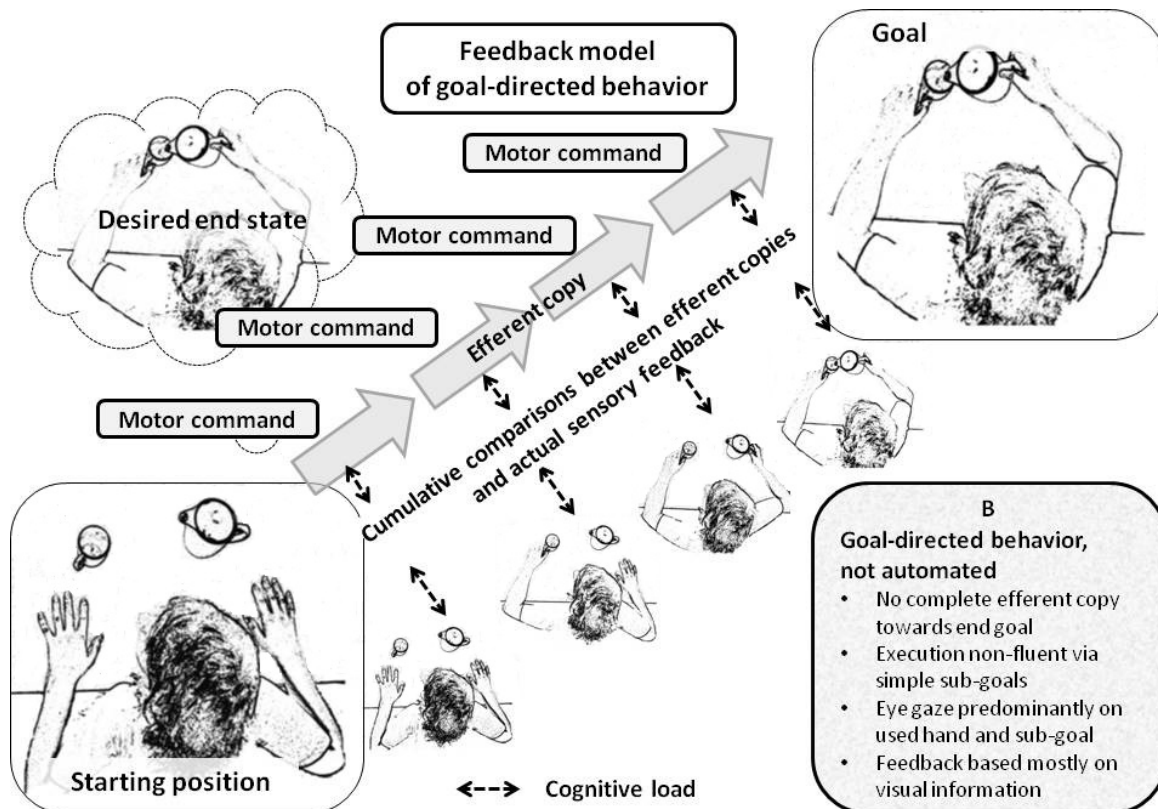
**Figure 5a (additional): Motor Imagery in typically developing children.**

*Schematic representation of a feedforward model of well learned goal-directed behavior. Before movement execution, the complete complex motor sequence is planned. Thus, an efference copy of the full motor sequence is thought to be formed allowing a fluent and efficient execution of the motor sequence with minimal cognitive control for ongoing feedback comparison to track and correct the motor sequence during execution. Explicit motor imagery is thought to provide a window on the efference copy without the actual motor sequence being executed.*



**Figure 5b (additional): Motor Imagery in children with an a-typical motor development (e.g. CP).**

*Schematic representation of a feedback approach of a goal-directed complex motor sequence that is not (fully) automated. At the start of movement execution, the first step of the complex behavioral sequence is planned and executed followed by the next step of the complex behavioral sequence and so forth. This results in a relatively slow execution of an iterative motor sequence with the necessity of a high amount of cognitive control for ongoing feedback and planning of stepwise sub-goals (see also Steenbergen and van der Kamp, 2004). Because there is no initial formation of a complete efference copy, motor imagery would be diminished and incomplete.*



In conclusion, we found that children with unilateral CP were able to engage in a 1<sup>st</sup> person kinematic approach to solve the HLJ task with respect to their less-affected hand. However, with respect to the affected hand these children seemed to rely on a visual rotation strategy to solve the task suggesting a diminished proprioception and less access to the body schema in children with unilateral CP when the affected hand involved. This suggests that children with CP could benefit from intervention based on visual training strategies.

### **Acknowledgement**

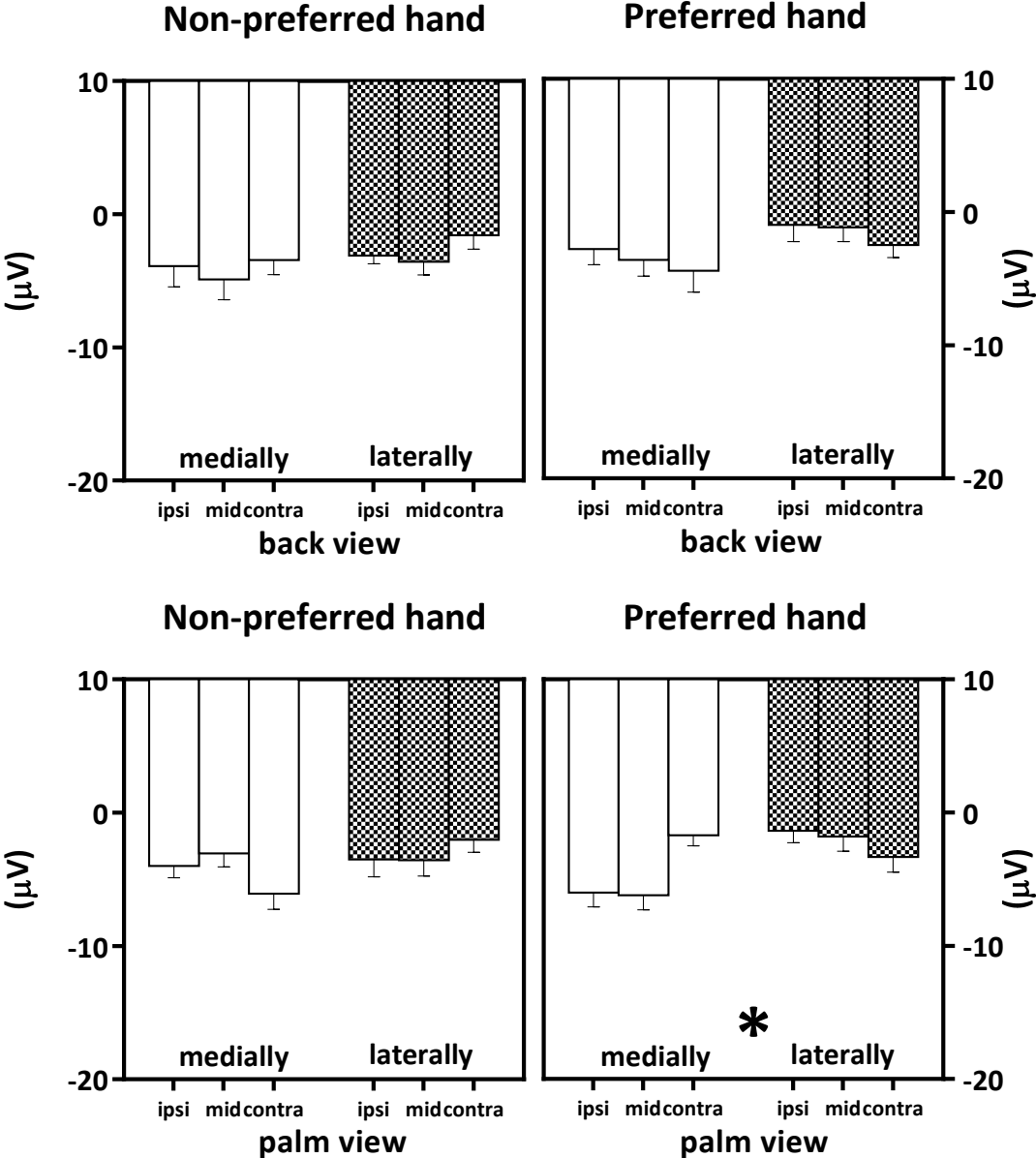
The research was funded by the Netherlands Scientific Research NWO Brain and Cognition program project nr: 433-09-215.

## References

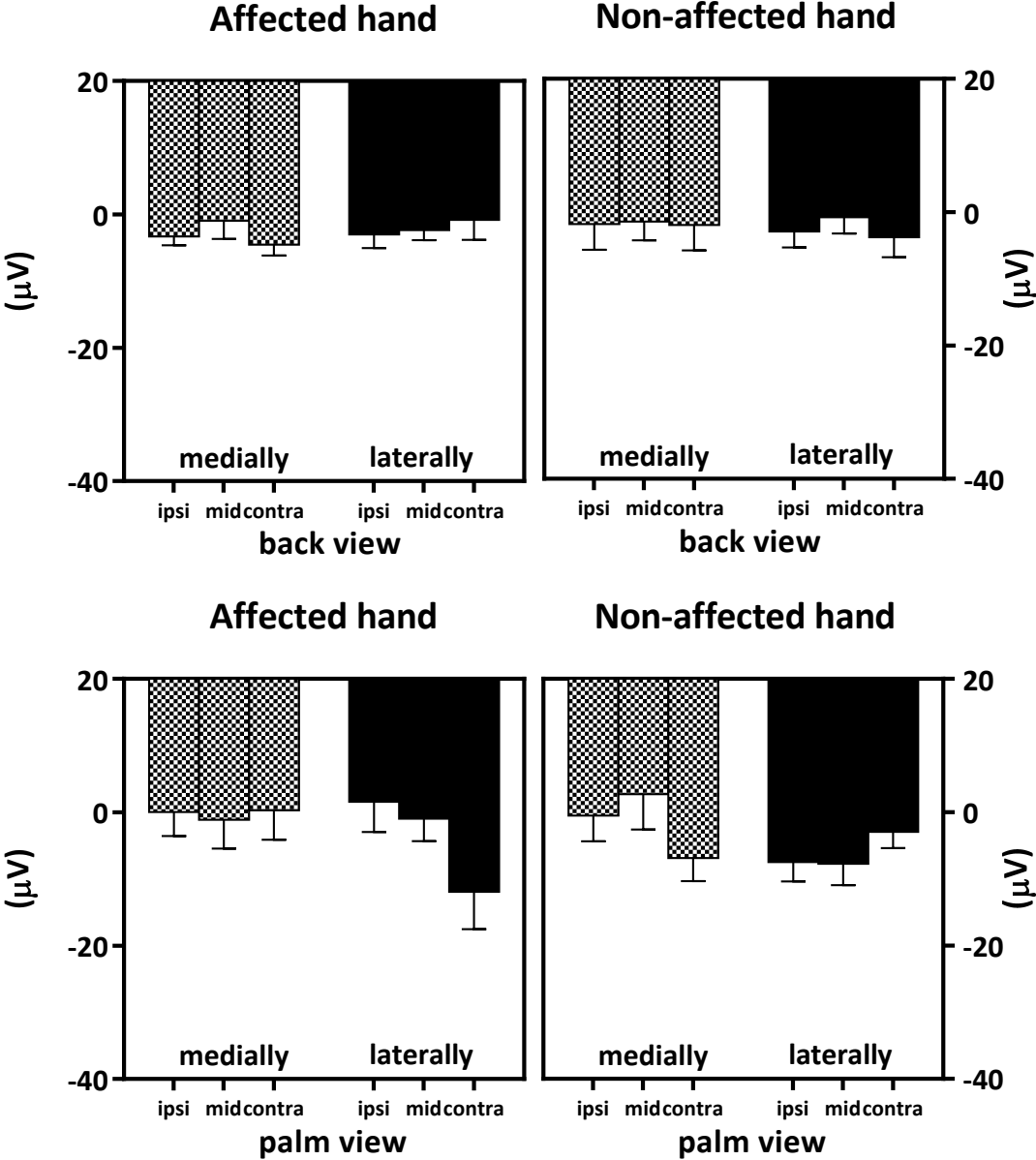
- [1] Rosenbaum P, Paneth N, Leviton A, Goldstein M, Bax M. A report: The definition and classification of cerebral palsy april 2006. *Dev Med Child Neurol* 2007; 49: 8-14.
- [2] Gordon AM, Bleyenheuft Y, Steenbergen B. Pathophysiology of impaired hand function in children with unilateral cerebral palsy. *Dev Med Child Neurol* 2013; 55: 32-37.
- [3] Steenbergen B, van der Kamp J. Control of prehension in hemiparetic cerebral palsy: similarities and differences between the ipsi- and contra-lesional sides of the body. *Dev Med Child Neurol* 2004; 46: 325–332.
- [4] Steenbergen B, Gordon AM. Activity limitation in hemiplegic cerebral palsy: evidence for disorders in motor planning (review). *Dev Med Child Neurol* 2006; 48: 780–83.
- [5] Steenbergen B, Jongbloed-Pereboom M, Spruijt S, Gordon AM. Impaired motor planning and motor imagery in children with cerebral palsy: Challenges for the future of pediatric rehabilitation. *Dev Med Child Neurol*; 2013: 55, 43-46.
- [6] Daprati E, Nico D, Sylvie Duval S, Lacquaniti F. Different motor imagery modes following brain damage. *Cortex*; 2010: 46, 1016-1030.
- [7] Shentona JT, Schwoebel J, Cosletta HB. Mental motor imagery and the body schema: evidence for proprioceptive dominance. *Neurosci Lett*; 2004: 370, 19–24.
- [8] Fontes PLB, Moura R, Haase VG. Evaluation of body representation in children with hemiplegic cerebral palsy: toward the development of a neuropsychological test battery. *Psychol Neurosci*; 2014: 7, 139 – 149.
- [9] Parsons LM Temporal and kinematic properties of motor behavior reflected in mentally simulated action. *J Exp Psychol Hum Percept Perform* 1994; 20: 709-30
- [10] Thayer ZC, Johnson BW. Cerebral processes during visuo-motor imagery of hands. *Psychophysiol* 2006; 43: 401–412.
- [11] Jongsma MLA, Meulenbroek RGJ, Okely J, Baas CM, van der Lubbe RHJ, Steenbergen B. Effects of hand orientation on motor imagery - Event related potentials suggest kinesthetic motor imagery to solve the hand laterality judgment task. *PLoS One* 2013; 8: e76515.
- [12] Ter Horst AC, Jongsma MLA, Janssen LK, van Lier R, Steenbergen B. Different mental rotation strategies reflected in the rotation related negativity. *Psychophysiol* 2012; 49: 566–573.
- [13] Steenbergen B, van Nimwegen M, Crajé C. Solving a mental rotation task in congenital hemiparesis: motor imagery versus visual imagery. *Neuropsychologia* 2007; 45: 3324–3328.
- [14] Williams J, Anderson V, Reddihough DS, Reid SM, Vijayakumar NV, Wilson PH. A comparison of motor imagery performance in children with spastic hemiplegia and developmental coordination disorder. *Journal of Clinical and Experimental Neuropsychol* 2011a; 33: 273–282.

- [15] Williams J, Reid SM, Reddihough DS, Anderson V. Motor imagery ability in children with congenital hemiplegia: Effect of lesion side and functional level. *Res Dev Disabil* 2011b; 32: 740–748.
- [16] Eimer M. Crossmodal links in spatial attention between vision, audition, and touch: evidence from event-related brain potentials. *Neuropsychol* 2001; 39:1292–1303
- [17] Van Elk M, Crajé C, Beeren MEGV, Steenbergen B, van Schie HT, Bekkering H. Neural evidence for impaired action selection in right hemiparetic cerebral palsy. *Front Neurol* 2010; 1: 1-7.
- [18] Johnson-Frey SH. Stimulation through simulation? Motor imagery and functional reorganization in hemiplegic stroke patients. *Brain Cogn* 2004; 55: 328–331.
- [19] American Encephalographic Society. Guidelines for standard electrode position nomenclature. *J Clin Neurophysiol* 1994; 11: 40-73.
- [20] Di Rienzo F, Collet C, Hoyek N, Guillot A. Impact of neurologic deficits on motor imagery: A systematic review of clinical evaluations. *Neuropsychol Rev*; 2014: 24, 116–147.
- [21] Crajé C, van Elk M, Beeren M, van Schie HT, Bekkering H, Steenbergen B. Compromised motor planning and motor imagery in right hemiparetic cerebral palsy. *Res Dev Disabil* 2010; 31: 1313–1322.
- [22] Lust JM, Geuze RH, Wijers AA, Wilson PH. An EEG study of mental rotation-related negativity in children with Developmental Coordination Disorder. *Child Care Health Dev* 2006; 32: 649–663.
- [23] Steenbergen B, Hulstijn W, de Vries A, Berger M. Bimanual movement coordination in spastic hemiparesis. *Exp Brain Res* 1996; 110: 91–98.
- [24] Sharma N, Pomeroy VM, Baron JC. (2006). Motor imagery: a backdoor to the motor system after stroke? *Stroke*, 37, 1941–1952.
- [25] Wilson PH, Thomas PR, Maruff P. Motor imagery training ameliorates motor clumsiness in children. *J Child Neurol* 2002; 17: 491–498.

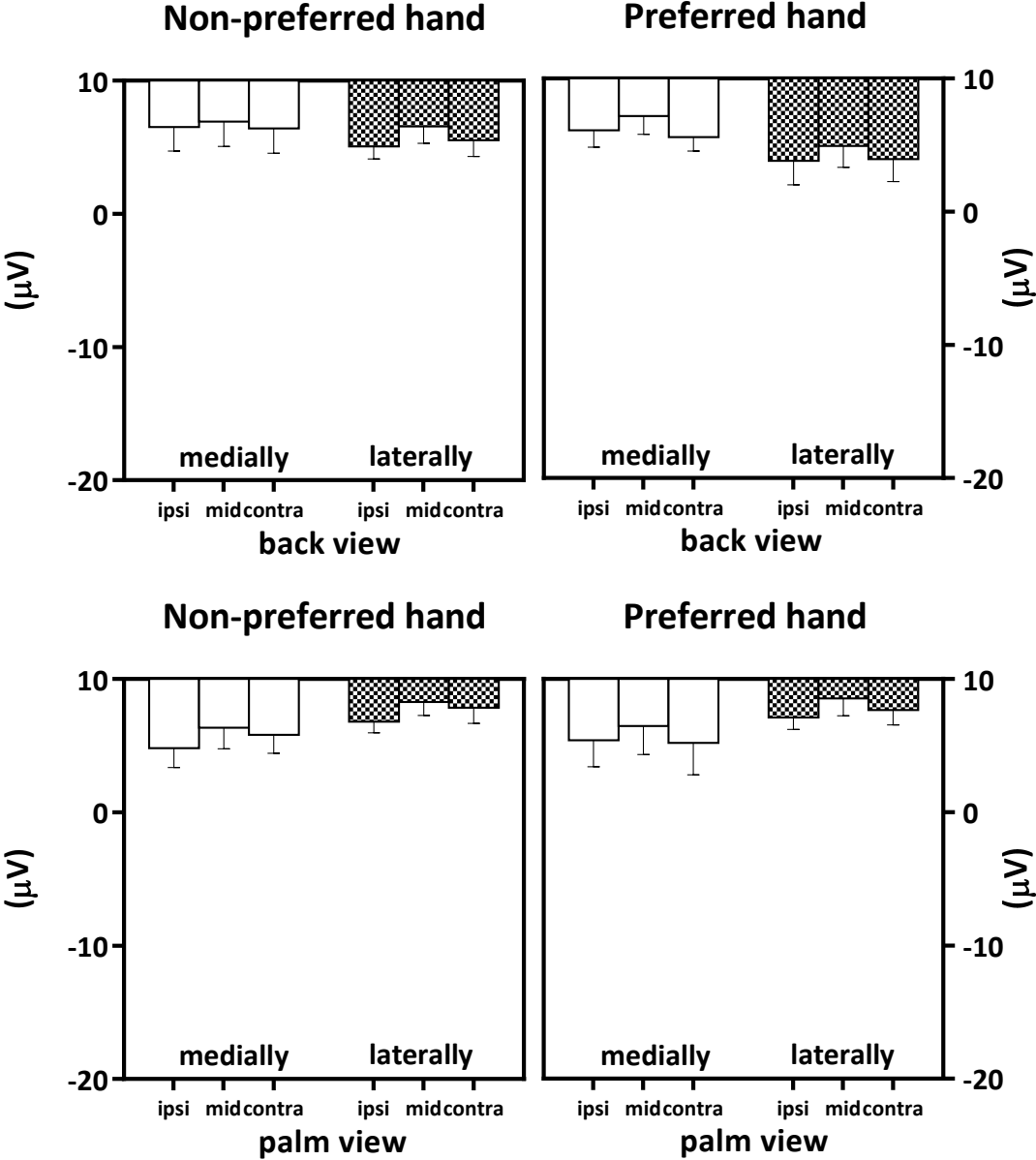
# N1 amplitude typically developing children



# N1 amplitude children with unilateral CP

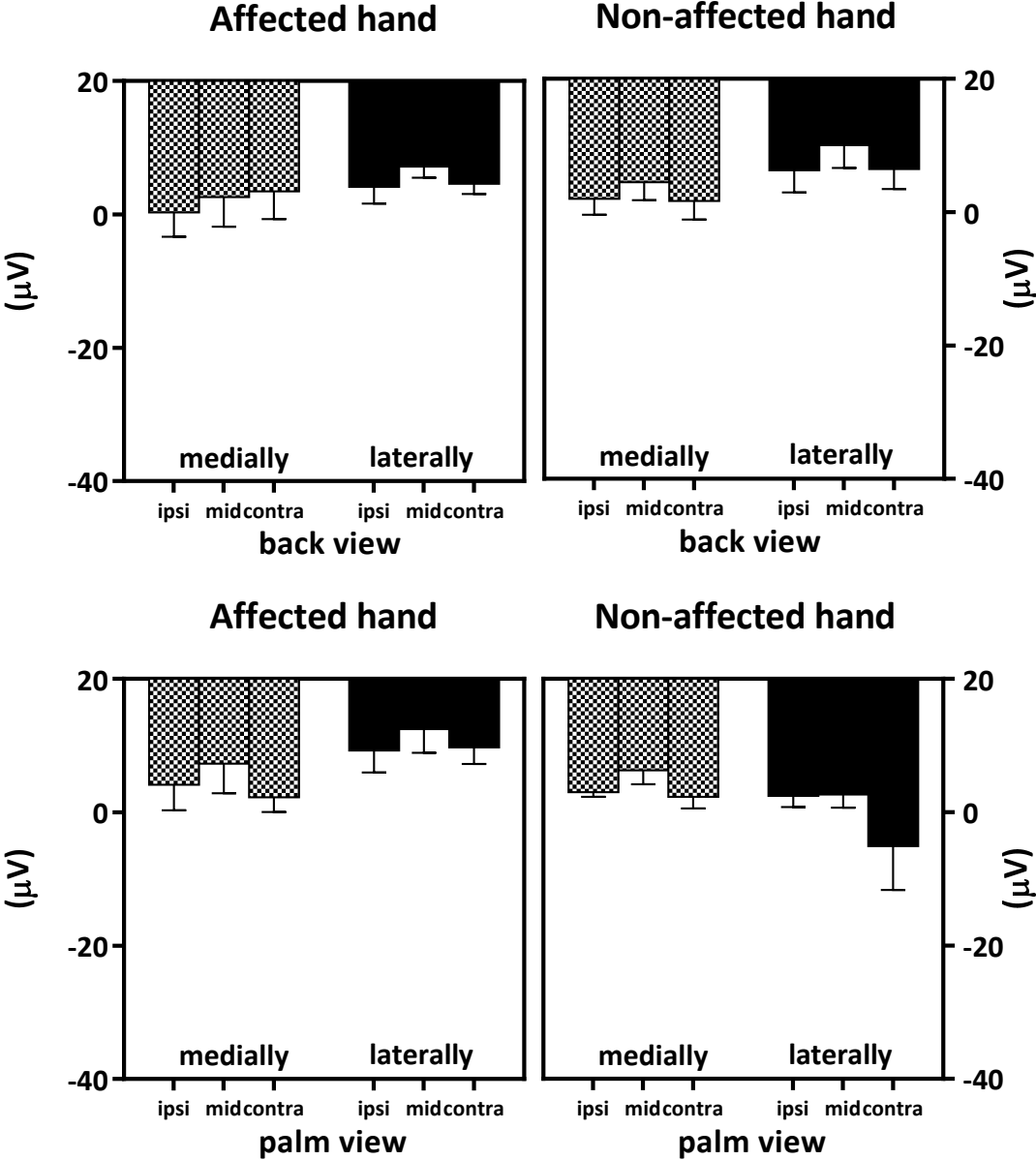


## P2 amplitude typically developing children

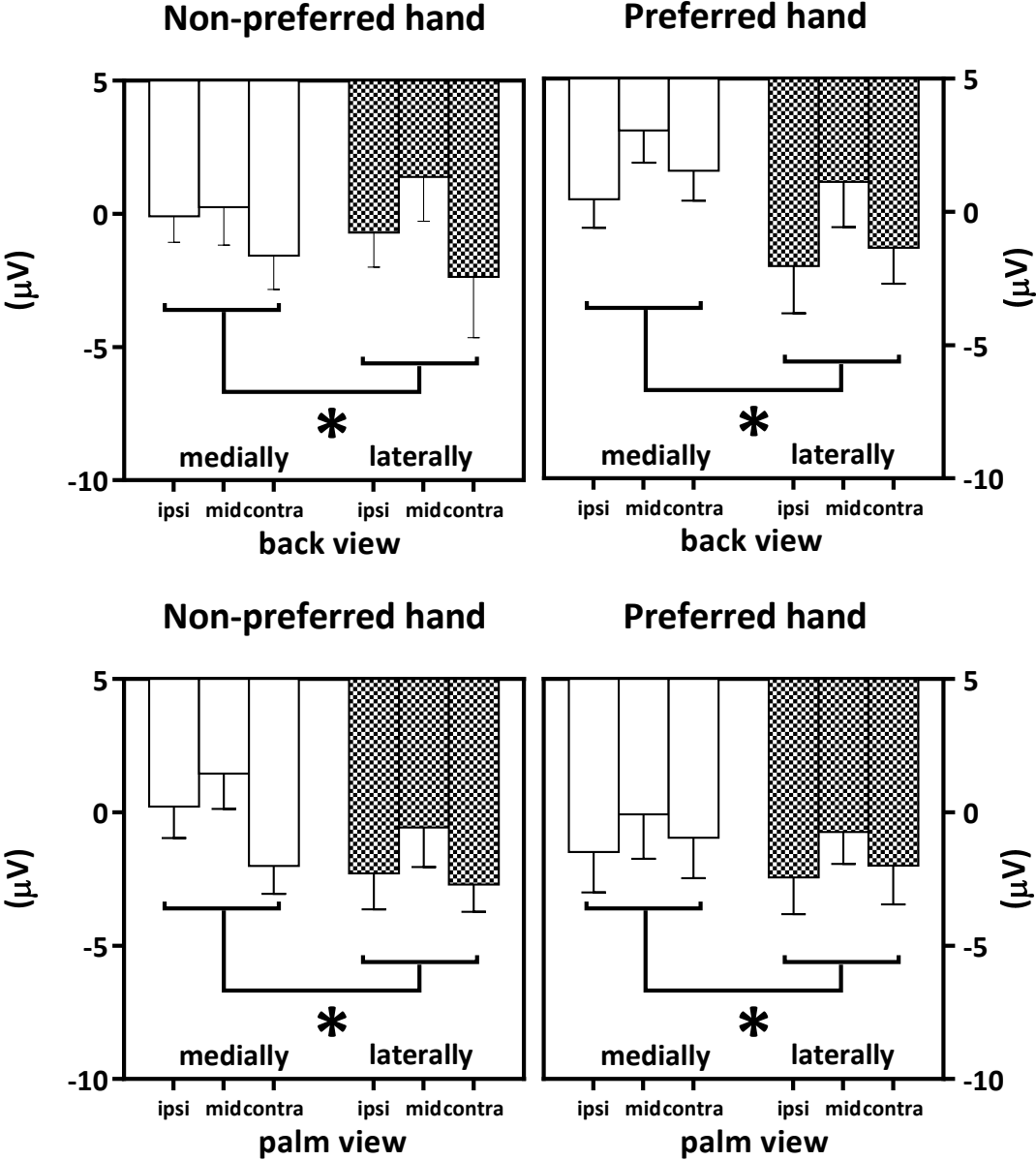




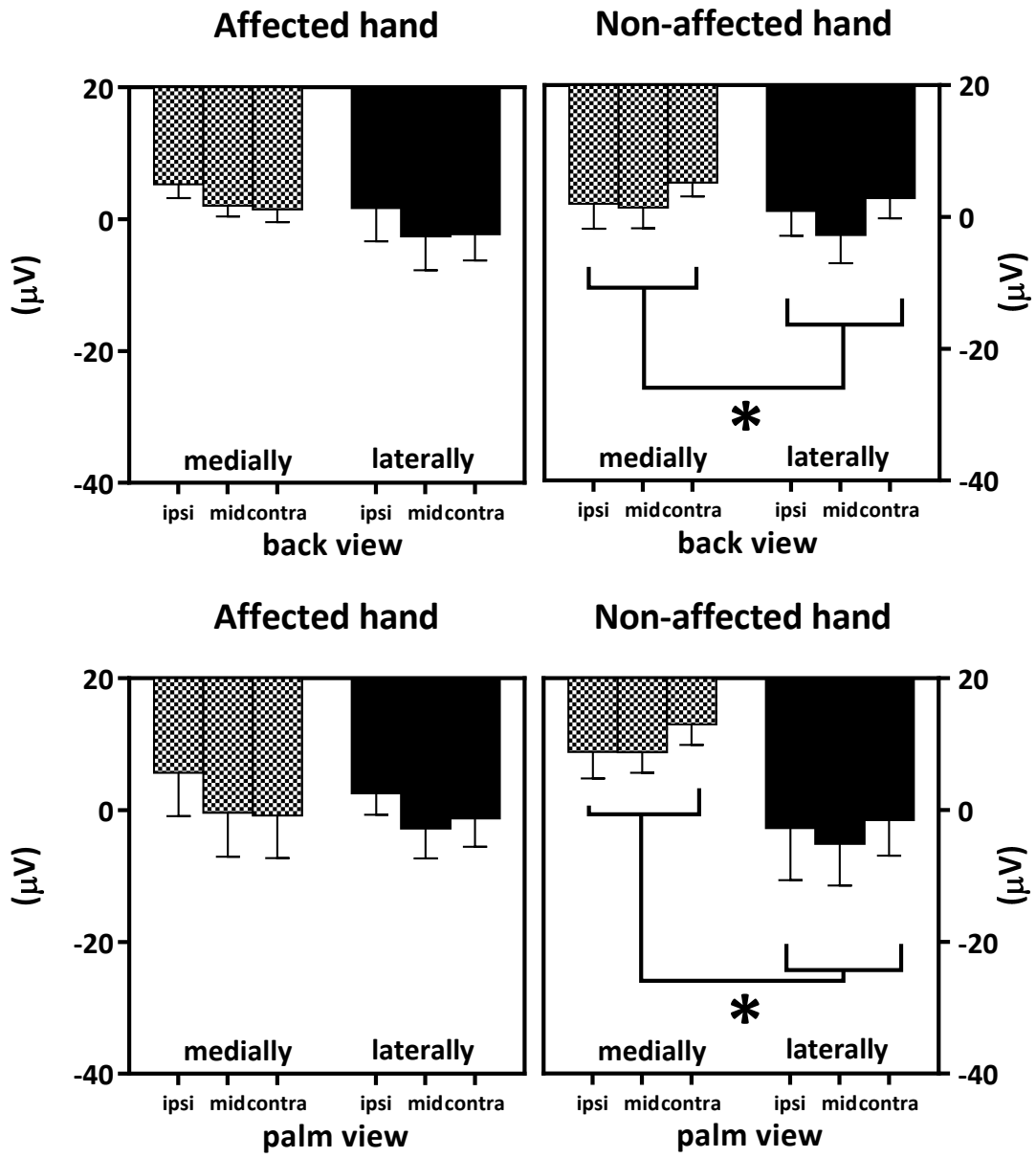
## P2 amplitude children with unilateral CP



# RRN amplitude typically developing children



# RRN amplitude children with unilateral CP



## **Legends**

### **Supplementary material 1a: N1 amplitude results of the group of typically developing (TD) children**

N1 (the mean voltage between 140-160 ms after stimulus presentation) is depicted for the TD group. Medially rotated stimuli are depicted with white bars whereas laterally rotated stimuli are depicted by dotted bars. RRNs are depicted for ipsi- mid- and contra-lateral parietal electrodes with respect to the depicted hand stimulus. The asterisk mark the significant effect ( $P < .05$ ).

### **Supplementary material 1b: N1 amplitude results of the group of children with unilateral Cerebral Palsy (CP)**

N1 (the mean voltage between 140-160 ms after stimulus presentation) is depicted for the CP group. Medially rotated stimuli are depicted with dotted bars whereas laterally rotated stimuli are depicted by black bars. RRNs are depicted for ipsi- mid- and contra-lateral parietal electrodes with respect to the depicted hand stimulus.

### **Supplementary material 2a: P2 amplitude results of the group of typically developing (TD) children**

P2 (the mean voltage between 200-220 ms after stimulus presentation) is depicted for the TD group. Medially rotated stimuli are depicted with white bars whereas laterally rotated stimuli are depicted by dotted bars. RRNs are depicted for ipsi- mid- and contra-lateral parietal electrodes with respect to the depicted hand stimulus.

### **Supplementary material 2b: P2 amplitude results of the group of children with unilateral Cerebral Palsy (CP)**

P2 (the mean voltage between 200-220 ms after stimulus presentation) is depicted for the CP group. Medially rotated stimuli are depicted with dotted bars whereas laterally rotated stimuli are depicted by black bars. RRNs are depicted for ipsi- mid- and contra-lateral parietal electrodes with respect to the depicted hand stimulus.

### **Supplementary material 3a: Rotation Related Negativity (RRN) results of the group of typically developing (TD) children**

RRN (the mean voltage between 350-400 ms after stimulus presentation) is depicted for the TD group. Medially rotated stimuli are depicted with white bars whereas laterally rotated stimuli are depicted by dotted bars. RRNs are depicted for ipsi- mid- and contra-lateral parietal electrodes with respect to the depicted hand stimulus. Asterisks mark the significances ( $P < .05$ ).

### **Supplementary material 3a: Rotation Related Negativity (RRN) results of the group of children with unilateral Cerebral Palsy (CP)**

RRN (the mean voltage between 350-400 ms after stimulus presentation) is depicted for the CP group. Medially rotated stimuli are depicted with dotted bars whereas laterally rotated stimuli are depicted by black bars. RRNs are depicted for ipsi- mid- and contra-lateral parietal electrodes with respect to the depicted hand stimulus. Asterisks mark the significances ( $P < .05$ ).

