Twenty weeks of computer-training improves sense of agency in children with spastic cerebral palsy

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\textbf{ABSTRACT}

Children with cerebral palsy (CP) show alteration of perceptual and cognitive abilities in addition to motor and sensory deficits, which may include altered sense of agency. The aim of this study was to evaluate whether 20 weeks of internet-based motor, perceptual and cognitive training enhances the ability of CP children to determine whether they or a computer are responsible for the movement of a visually observed object. 40 CP children (8–16 years) were divided into a training (n:20) and control group (n:20). The training group trained 30 min each day for 20 weeks. The ability of the children to judge whether they themselves or a computer were responsible for moving an object on a computer screen was tested before and after the 20-week period. Furthermore, we included a healthy age-matched group to determine a normal functional level of performance. Our results showed a significantly larger increase in the number of correct subjective reporting for the training group (p < 0.001). In accordance with this, the training group was also less fooled by computer-induced movements given by a decreased curvature which indicated a compensatory motor strategy when drawing the line to hit the target following the training than the control group (p = 0.018). These findings suggest that sense of agency may be altered, and that training of sense of agency may help to increase the outcome of training programmes in children with CP.

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1. Introduction

Although cerebral palsy (CP) is commonly described as a non-progressive disorder of normal sensory–motor development, new research has emphasized that CP also involves alteration of perception and cognitive abilities depending on the site, extent and time during development of the lesion\textsuperscript{(Bottcher, 2010; vanderWeel, vanderMeer, & Lee, 1996). Recent findings have shown that a population of children with spastic CP has significantly larger problems than healthy children in determining whether an observed movement is caused by themselves or a computer\textsuperscript{(Ritterband-Rosenbaum et al., 2011). Decreased kinesthetic perception of limb position\textsuperscript{(Riquelme & Montoya, 2010; Wingert, Burton, Sinclair, Brunstrom, & Damiano, 2009), disturbances of perception and cognition\textsuperscript{(Bottcher, 2010; vanderWeel et al., 1996) and a general lower score of own physical competence have also been reported in children with CP\textsuperscript{(Russo et al., 2008). Ritterband-Rosenbaum}}

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et al. (2011) consequently suggested that CP children have a reduced ability to interpret the sensory information caused by their own motor behavior and correctly define the proper agent of a movement. This ability is generally referred to as sense of agency (Farrer & Frith, 2002; Nielsen, 1963).

A key element of the sense of agency is the ability to differentiate self-produced movements from movements originating from the external environment (Farrer & Frith, 2002; Franck et al., 2001; Sirigu, Daprat, Pradat-Diehl, Franck, & Jeannerod, 1999). When the sensory modalities provide feedback caused by the movement, which matches with the intention of the planned movement, participants experience agency of the specific movement. On the other hand, when there is a discrepancy between the actual sensory feedback and the intended sensory feedback, the sense of agency disappears (Farrer & Frith, 2002; Geiregoff & Jeannerod, 1998; Synofzik, Thier, & Lindner, 2006).

In the present study we hypothesised that CP children’s capability to correctly perceiving their own movement, can be improved with training, which stimulates cognitive processes and motor control. This could be of great importance for successful rehabilitation of normal daily function for children with CP as well as other patients with brain damage (see e.g. Oliver Sachs for first person observation; Sachs, 1984).

2. Material and methods

2.1. Ethics approval

The research was approved by the local ethics committee (protocol number: H-B-2009-17) and the study was conducted in accordance with the declaration of Helsinki guidelines. All participants were given written and verbal introductions regarding the experiment. All the children, parents and/or legal guardians provided a written consent form before data collection.

2.2. Participants

We designed a controlled longitudinal study, which consisted of a total of 40 CP children divided in a training group CP_{Train} and a control group CP_{Con} comprised of 20 children in each group. We also included a healthy age-matched group of 65 children (average age: 11.6 ± 2.2, 37 boys/28 girls). This group only completed the experiment in a single session. We used the responses to define the average level of normal performance for the specific tasks. Parts of the data for these healthy control children have been published in Ritterband-Rosenbaum et al. (2011).

Table 1 shows data for the two CP groups including sex and mean age for the groups. We selected the CP children on the basis of medical records in order to create groups which were clinically homogenous based on the CP children’s Gross Motor Function Classification Score (GMFCS-level) (Palisano et al., 1997), their Manual Ability Classification System (MACS) (Eliasson et al., 2006) and their lesion site. The children also performed a Test of Visual Perception Skills (TVPS) consisting of 7 different visual perception tests. Overall performance in the tests is given by the sum of each individual test’s score scaled according to the normative data for the child’s age group and a normalisation score (Gardner, 1996). It was not possible to obtain TVPS data from 1 child in the CP_{Train} and 7 children in the CP_{Con} group and MACS and GMFCS for 5 children in the CP_{Con} group for practical reasons, but for the remaining children there was no significant difference in the scoring between the two groups. There was no significant age-difference between the two groups (t-test, p = 0.5). There was no statistical difference between the two groups in the number of children with lesion in the right or left hemisphere (p = 0.1).

2.3. Apparatus and procedure

The experimental task consisted of a relatively short session, where the participants were seated at a table performing reaching movements by moving an object on a pen-tablet to a cued target in a straight ballistic movement as quickly and precisely as possible (cf. Fig. 1). All participants used their preferred hand to ensure that they were all able to perform the task. The actual arm movement was hidden, but participants were capable of following the movement by looking at a monitor in front of them. After each movement the participants had to verbally evaluate whether they perceived the

<table>
<thead>
<tr>
<th>Sex</th>
<th>Age</th>
<th>Handedness</th>
<th>Diagnosis (right/left)</th>
<th>GMFCS</th>
<th>MACS</th>
<th>TVPS scaled score</th>
<th>TVPS norm score</th>
</tr>
</thead>
<tbody>
<tr>
<td>CP_{Train} boys</td>
<td>11.1 ± 2.1</td>
<td>12 right</td>
<td>20 Hemiplegia 8 Rt/11 Lt (1)</td>
<td>19 I</td>
<td>11 I</td>
<td>67.1 ± 21.1</td>
<td>97.7 ± 15.2</td>
</tr>
<tr>
<td>CP_{Train} girls</td>
<td>12.0 ± 2.6</td>
<td>10 right</td>
<td>20 Hemiplegia 11 Rt/4 Lt (5)</td>
<td>15 I</td>
<td>12 I</td>
<td>70.0 ± 19.6 (7)</td>
<td>99.8 ± 14.0 (7)</td>
</tr>
</tbody>
</table>

The table describes the data for the two CP-groups. The diagnosis also includes the lesion sites when it was possible to obtain the information. GMFCS = gross motor function classification scale. MACS = manual assessment classification score. TVPS = Test of visual perception skills. This score is given as the scaled score and the normalized score according to the age of the children. Numbers in parenthesis are missing data.
movement of the visual object as being made by themselves or by a computer, which randomly manipulated the moving object by producing trajectories. The trajectories diverted either 10° or 15° away from the target. During the experiment, other manipulations were introduced, however for analysis purpose, we chose only to use those two deviations as our previous study has shown that these manipulations gave rise to significant differences between CP children and their healthy peers, and because only those manipulations corresponded to report rates close to chance level for healthy participants (Ritterband-Rosenbaum et al., 2011).

All participants were encouraged to answer as fast as possible in order not to allow too long a time for retrospective consideration of their own motor performance and the visual feedback. Furthermore, all participants were informed prior to the experiment that they were only responsible for completing the movements in some of the trials. For further details of the experimental setup see (Ritterband-Rosenbaum et al., 2011).

2.4. Data collection and analysis

Behavioral data containing the \(X_{\text{pen}}, Y_{\text{pen}}\) coordinates for each individual movement produced by the pen on the tablet and the \(X_{\text{screen}}, Y_{\text{screen}}\) coordinates of the visual object on the screen was also recorded for off-line analysis. The data coordinates were sampled at 60 Hz. All the data containing distance measurements are normalized to the size of the pen-tablet.

Only information on line-drawings produced in completed movements were stored for later analysis, thereby excluding data information regarding unfinished lines due to lifting of the pen or drawing out of area on the pen-tablet. For further data analysis we have pooled 10° and 15° manipulations and excluded all other trials.

Only data from between 20% and 70% of the vertical display screen was used to avoid data contamination which could appear when the participant placed the pen on the table to “pick up” the visual object and when they finished the lines. Furthermore, we excluded trials where the curvature exceeded three standard deviations compared to the average of the line produced by the subject within the specific manipulations of the visual object’s trajectory or if the \(X_{\text{pen}}, Y_{\text{pen}}\) coordinates for individual trials were insufficient. This resulted in a small percentage of trials being excluded from \(\text{CP}_{\text{Train}}\) (3.3%) and \(\text{CP}_{\text{Con}}\) (5.5%).

The behavioral kinematic and subjective measurements were given by:

1) Hit-distance: corresponds to the distance between the hit-target and the end position of the \(X_{\text{pen}}, Y_{\text{pen}}\) coordinates (mm) (1).
2) Time: indicates the time to complete the movement (mm) (2).
3) Curvature (C); the curvature is given by:

\[ C = \frac{x'y'' - y'x''}{(x^2 + y^2)^{3/2}} \]

We used the absolute summed values given by the \(C\) for each data point (mm\(^{-1}\)). This value illustrates how much the participants tried to compensate for the deviation of the moving object during the course of the trajectory to the target (cf. Fig. 1).

4) Subjective assessment of reporting “Yes” or “No” of being responsible for the movement.
2.5. Training

The training was conducted over a period of 20 weeks. During this period the CPTrain group trained for at least 30 min daily using the Internet-based home training system ‘Move It to Improve It’ (MiTii) (Bilde et al., 2011), and the CPCon continued their regular daily activities. The CP children in the control group were specifically instructed to follow their normal habits in relation to their physical activity level and their computer habits.

MiTii (MiTii-development, Copenhagen, Denmark) is a web-based training program for CP children. It is designed to combine cognitive and motor challenges in order to train cognitive, perceptual and motor abilities at the same time. It consists of a number of training modules in which the child has to analyze visual information, solve a cognitive problem (ex. solving mathematical question, remembering a sequence of images, etc.) and respond with a motor act (ex. blowing up a balloon with the correct answer to a math-question, dragging the correct images from a variety of options, maneuvering through a cave without hitting the wall, etc.) The core of the system is that the computer program identifies the movements of the child from video images sampled from a simple web-camera attached to the computer. The child may control the computer-program by his or her own free movements as the webcam detects motion of a green band which may be placed around the hands, arms, shoulder, head, wrist or legs of the child. The training modules are customized to fulfill each CP child’s level of difficulty by adjusting the complexity of the specific tasks (additional repetitions of the task, extra objects or more difficult recognizable objects within the task) to optimize training progresses and to ensure each training session lasted about 30 min each day in the 20-week period. The specific modules for each training program were combined according to each child’s specific perceptual and motor baseline scores (Bilde et al., 2011).

2.6. Statistics

We used 2-way-repeated measures analysis of variance to determine if the behavioral experimental data varied across groups for pre versus post training. The population mean, standard deviation and the Standard Error of the Mean (SEM) of observation within the two groups were calculated for each individual motor parameter (hit distance, time, curvature) as well as the subjective assessment score. In case of interactions between the factors we proceeded with a post hoc analysis for multiple comparisons using Bonferroni correction. The statistical differences are therefore reported as the results from the post hoc tests.

For comparison between the subjective assessments we used \( \chi^2 \) for each individual movement type. The level of significance was set to \( p < 0.05 \).

The graphs are displayed as averages across groups with 1 intersubject SEM.

3. Results

Before the training intervention, children in the CPTrain group on average reported that they were responsible for the movement in \( 48 \pm 5.2\% \) of trials whereas the CPCon had an average score of \( 48 \pm 5.6\% \). After training the children in the CPTrain group significantly more frequently reported correctly whether they or the computer was responsible for the movement \( (31.5 \pm 4.8, \text{ cf. Fig. 2}) (p = 0.005) \). A similar change was not observed in the CPCon group \( (37.5 \pm 4.3) (p = 0.9) \). Furthermore, we found a significant difference between the two groups after the training intervention giving an improved subjective reporting rate.

![Fig. 2. The sense of agency. The cognitive perception of being responsible for the movements is given by the percentage of the verbal reporting rate for Pre and Post training intervention. Black circles are CPTrain and grey circles are CPCon. Error bars: 1 intersubject SE. The dotted line represents the level of healthy age-matched children. Asterisk indicate a significant difference of \( p < 0.001 \).](image)
Fig. 3. Kinematics of the movements for the two groups. A–C illustrates the average results for the different kinematic properties of the combined 10° and 15° visual manipulation divided into the two groups of CP children (black circles: CP_train, grey circles: CP_con) during pre-intervention and post-intervention experiments. Error bars: 1 intersubject SE. The dotted lines indicate data for an age-matched healthy group of children to show the normal level of performance. The asterisks indicate significant value of \( p < 0.05 \). (A) The graph displays the average hit distance (mm) for CP_train and CP_con, respectively. (B) The graph depicts the averaged time (ms) it takes to complete the movement manipulations for the two groups. (C) The graph illustrates the curvature (mm \(^{-1}\)) for the groups. The higher the curvature is, the greater the CP children have tried to compensate for the introduced computer manipulation of the visual object. We did not find any significant difference for the CP_con.
for the CP_train group vs. the CP_con group (p < 0.001). Following the training the children in the CP_train group had a frequency of correct reports, which was similar to that of healthy age-matched children (Fig. 2; horizontal line) (p = 0.9).

The kinematics of the performed movements is presented in Fig. 3. There were no significant changes in the ability of the children to hit the target before and after the 20 weeks intervention in any of the two groups (Fig. 3A), but the time to complete the movement decreased significantly for the two groups from prior and after the intervention (Fig. 3B) of 60% and 72% for the CP_train and CP_con groups, respectively (F(1,38) = 37.3; p < 0.001). The ability of the children to correct the computer-induced deviation of the object away from the target is given by how far and for how long the line deviated from the straight line connecting the starting point and the target. This was calculated as the curve of the line (see Section 2). As can be seen from Fig. 3C the children in the CP_train group showed a significantly smaller curve following the training (p = 0.02). In contrast, no difference was found for children in the CP_con and a two-way ANOVA test revealed a statistically significant difference in the test prior and after the intervention between the two groups (F(1,38) = 6.061; p < 0.05).

The change in subjective report of agency showed a weak correlation with the change in curvature and time for the CP_train group (r = 0.3, p = 0.1), but not for the CP_con group (r = 0.09, p = 0.7) post the training intervention. The same correlations were found for the time the two groups used to complete the movements.

Furthermore, we did not find any statistical difference in the subjective reporting rates between the two groups related to their lesion site (left or right) (F(1,592) = 1.541, p = 0.2). Nor did we find any correlation between the TVPS and the subjective reporting for the two groups (correlation coefficients below 0.05).

4. Discussion

We have shown in this study that children with CP improve their ability to determine whether they themselves or a computer are responsible for the movement of an observed object following 20 weeks of interactive computer training designed to strengthen sensory–motor interaction. This was also reflected behaviorally as a reduced compensatory reaction when the computer introduced a deviation of the observed object. We propose that the training induced an improved sense of agency and that this may be of importance for rehabilitation of functional capacities in children with CP. The improved ability of the children in the present study to report when they themselves were responsible for the movement likely reflects an improved ability to couple their intention of movement with the resulting consequences in the form of a moving object on the computer screen. In this sense it seems reasonable to assume that the children became better at monitoring and comparing their own intentions and the sensory feedback evoked by their movements and adequately discern any discrepancy between them. This ability is usually described in the literature as sense agency and appears to depend on processing of visual, proprioceptive and motor command information in the parietal cortex (Daprati et al., 1997; Daprati, Nico, Duval, & Lacquaniti, 2010; Sato & Yasuda, 2005; Yomogida et al., 2010).

Agency is an important factor for our self-consciousness and the ability to control our movements and interact appropriately with the environment. The computer-model we used in our experiment to investigate sense of agency was reported originally by Ritterband-Rosenbaum et al. (2011), and was inspired by Nielsen (1963). Newer studies have adapted the paradigm in order to study the underlying network of sense of agency in healthy and brain-damaged participants. In these experiments participants have been presented video-images showing either their own hand or the hand of an experimenter performing movements that are either congruent or incongruent with the participants own movements (Daprati et al., 1997, 2010; Sato & Yasuda, 2005; Sirigu et al., 1999; Yomogida et al., 2010). In all of the studies, participants were to evaluate if they were responsible for the observed movement or not. The results have shown that lesions in especially the (right) parietal regions cause a decrease in correct judgment of the proper agent of a movement. We were not able to obtain imaging data of the CP children’s lesion, but their clinical symptoms indicated that lesions in the left and right hemisphere were almost equally represented in the study. There was no difference in the results for children with lesions in the left or right hemisphere, but this may be explained by the low number of subjects. As discussed by Ritterband-Rosenbaum et al. (2011) the clinical data indicate that all children suffered from reduced visual processing in addition to their motor symptoms. This would go along with lesions that involve areas in the posterior parietal cortex. Though, we did not find this in the present data.

Children in both the training and control group used less time to perform the movements on the second test. This may be due either to the fact that they performed the test a second time or that they had become 20 weeks older. Though, the improvement was found to be significantly better for the CP_train group compared to the CP_con group. The change in sense of agency in the training group thus had no measurable effect on the speed of the movement and vice versa. In contrast, the curvature of the movement, which is an indicator of the compensatory reaction of the children when the object on the computer screen deviated from the expected trajectory, was reduced only in the training group and a clear correlation with the reported sense of agency was observed. We take this as behavioral evidence that the children were less easy to ‘fool’ by the sudden change in the trajectory of the object induced by the computer following the training. In this relation it is of importance that the speed of the movement changed to the same extent in both groups since it might otherwise be argued that faster performance of the movement would make it more difficult for the children to detect the perturbation induced by the computer. However, in that case children in both groups should have shown an equal change in curvature also.
It is not surprising that the children in the training group improved their ability to correctly judge whether they or the computer was responsible for the movements, since the exercises that they trained daily for the 20-week period were designed to strengthen motor actions based on visual information of the consequences of their own movement on a computer screen (Bilde et al., 2011). There was thus a very good correspondence between the training and the test that we used for evaluation of sense of agency. However, this also raises the question as to whether the observed training effects can be transferred to other tasks, which are not computer-based. The observation by Bilde et al. (2011) that children do appear to improve their function also on more global functional scores following the 20 weeks training program suggests that this may be the case (Bilde et al, 2011). Consistent with this a few pilot studies, which have applied virtual reality computer interfaces as a rehabilitation source for training children with CP, have shown improvement of general functions as self-efficacy and motor performance after two times 90 min training/week during 4 weeks (Reid, 2002) and 9 h training during 3 weeks (Qiu et al., 2009). However, more specific studies will be necessary to determine to what an extent improved sense of agency in a computer-test such as ours may be transferred to more daily functional tasks and thus also be of importance for more general improvements in functional ability in children with CP following rehabilitation interventions.

5. Conclusion

We conclude that sense of agency can be improved in brain lesioned children after intense cognitive and motor control training. We propose that it should be considered that reduced sense of agency may have consequences for the functional abilities of patients and that strengthening of the sense of agency may be an important goal of rehabilitation strategies.

Authors’ contributions

ARR, MSC and JBN participated in the discussion and design of the experiment. ARR carried out all the experimental work. Data analysis was done by ARR and JBN. ARR and JBN wrote the manuscript. All authors read and approved the final manuscript.

Competing interest

The authors declare that they have no conflict of interest.

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