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Deficit in implicit motor sequence learning among children and adolescents with spastic Cerebral Palsy

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ABSTRACT

Skill learning (SL) is learning as a result of repeated exposure and practice, which encompasses independent explicit (response to instructions) and implicit (response to hidden regularities) processes. Little is known about the effects of developmental disorders, such as Cerebral Palsy (CP), on the ability to acquire new skills. We compared performance of CP and typically developing (TD) children and adolescents in completing the serial reaction time (SRT) task, which is a motor sequence learning task, and examined the impact of various factors on this performance as indicative of the ability to acquire motor skills. While both groups improved in performance, participants with CP were significantly slower than TD controls and did not learn the implicit sequence. Our results indicate that SL in children and adolescents with CP is qualitatively and quantitatively different than that of their peers. Understanding the unique aspects of SL in children and adolescents with CP might help plan appropriate and efficient interventions.

1. Introduction

The acquisition of motor skills is an essential component of development for all children (Haibach, Reid, & Collier, 2011). Motor skill learning (SL) refers to the cognitive process by which movements are executed more quickly and accurately with practice (Willingham, 1998). There is abundant and strong evidence for two separate motor learning systems in the brain: an explicit system that relies on strategies like instructions, feedback and demonstrations to generate performance while the process can be verbally described, and an implicit system that generates skilled motor performance unintentionally and often without awareness (Reder, Park, & Kieffaber, 2009; Squire, 2004).

The evidence supports the notion that the two systems are operating independently, by different neural networks, in the normal brain while acquiring a skill. A crucial anatomic difference between the two is the involvement of the medial temporal lobe (MTL) structures (i.e. the hippocampus and surrounding structures) in the explicit network and extra-MTL structures in the implicit system (Reder et al., 2009).

A simple task that has been thoroughly studied as a model of implicit skill acquisition is the serial reaction time (SRT) task, first reported by Nissen and Bullemer (1987). The task requires learning a sequence of motor responses to visual cues. Typically, four cue locations are shown on a computer screen with four possible keyboard button responses aligned underneath. Participants are asked to produce a motor response to a concealed *repeated sequence* presented in different spatial locations on a computer screen using corresponding buttons. After several *repeated sequence* blocks a *new sequence* is

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presented, followed by the *repeated sequence* once again at the end of the task. A gradual reduction in reaction time (RT) that takes place across the sequential blocks provides a measure of participants' mastering of the task – what Ferraro, Balota, and Connor (1993) termed "generalized skill". However, a more specific measure of skill acquisition is obtained by contrasting RT's for the *repeated sequence* against those of the *new sequence*. If following the replacement of the *new sequence*, participants implicitly continue to anticipate that the visual cues will occur at the same positions as in the *repeated sequence*, an implicit learning of the sequence has occurred (Vakil, Kahan, Huberman, & Osimani, 2000). This anticipation will inflate RT's in the *new sequence* trail, increasing the difference between responses to the *repeated* and *new* sequences. Tacit knowledge of the repeating sequence occurs even when participants are unaware of the existence of the sequence, and learning occurs at a normal rate in patients with memory disorders (Nissen & Bullemer, 1987). Studies of implicit learning patterns evaluating the SRT task in typically developing (TD) children showed that beyond typical patterns of decreasing reaction time throughout the task blocks and delays after introducing the new sequence, overall response time also decreased throughout childhood and adolescence (Janacsek, Fiser, & Nemeth, 2012).

Performance on SRT task has also been reported in children with atypical development, including children with autism (Barnes et al., 2008), ADHD (Barnes, Howard, Howard, Kenealy, & Vaidya, 2010), Williams and Down syndromes (Vicari, Verucci, & Carlesimo, 2007), and in children with structural brain anomalies involving the cerebellum (Vakil et al., 2000) and basal ganglia (Mayor-Dubois, Maeder, Zesiger, & Roulet-Perez, 2010), however findings were fairly inconsistent partially due to the different SRT methodologies used.

Cerebral Palsy (CP) is an umbrella term that describes a group of heterogeneous movement and posture disorders with varying etiologies and severity, attributed to non-progressive brain disturbances. These motor disorders are often accompanied by impairments in sensation, cognition, perception, and behavior (Bax et al., 2005).

As a result of motor disorders and accompanying impairments, children with CP manifest difficulties in executing various activities of daily living (ADL's) (Fauconnier et al., 2009). As with children who do not have neurological motor impairments, children with CP need to learn these activities in order to adequately function in the real world. Although visible motor difficulties like spasticity and limited range of motion are defining characteristics of the impairment in CP, it has been recently acknowledged that higher-order motor planning and motor learning deficiencies also exist (Bar-Haim et al., 2010; Steenbergen & Gordon, 2006). Considering intensive efforts invested in teaching motor skills to children with CP with only limited results (Liptak & Accardo, 2004), it has been suggested that the implicit system might offer a better avenue for improvement (Steenbergen, van der Kamp, Verneau, Jongbloed-Pereboom, & Masters, 2010). This necessitates investigation of the implicit system in children with CP.

Implicit forms of motor learning in this population have seldom been studied. In a series of experiments, Gagliardi, Tavano, Turconi, Pozzoli, and Borgatti (2011) and Gagliardi, Tavano, Turconi, and Borgatti (2013) examined SL in children with CP using the Corsi block test. Overall, compared to TD children, children with CP were less accurate and needed more attempts to learn. SL skills were unrelated to CP severity as defined by the Gross Motor Function Classification System (GMFCS) level (Palisano et al., 1997). In the follow up study, Gagliardi et al. (2013) found that SL skills are independent of age in TD and CP children. While these studies demonstrated how children with CP learn visuo-spatial sequences, their conclusions about SL abilities of children with CP should be considered with caution since they did not address the implicit learning component underlying their performance. This differentiation is crucial for understanding the cognitive processes of motor SL and can be achieved by using a more sensitive task, such as the SRT.

General intellectual abilities, measured as IQ, correlate with performance on explicit learning tasks but not with performance on implicit SL task when examined among TD children (Kaufman et al., 2010). Only a few studies addressed this relationship among children with developmental disabilities. For example, when assessing the relationship between non-verbal intelligence and performance on the SRT task in children with Down and Williams syndromes, Vicari et al. (2007) found IQ was independent of performance on the SRT task.

Given the importance of motor SL for children, the extensive clinical investment in this area for children and adolescents with CP yielding only moderate results, and the lack of knowledge about the performance of the implicit system in this population, our goal was to examine the explicit and the implicit components of motor SL among children and adolescents with CP compared to their TD peers, using the SRT task. In addition, we evaluated whether performance in the SRT task was related to individual differences such as age, cognitive ability and level of motor impairment.

2. Methods

2.1. Participants

Twenty-two children and adolescents (9 boys) diagnosed with spastic CP, aged 9–20 years (*M* = 13 y 2 mo, SD = 3 y 7 mo) and 23 TD controls (11 boys) aged 9–18 years (*M* = 13 y 1 mo, SD = 3 y 6 mo) volunteered to participate in the study. The participants were recruited from several mainstream schools, through special education schools and from the Department of Pediatric Rehabilitation in central Israel. All participants were tested in a quiet room either at their school or at the rehabilitation unit.

Inclusion criteria were a minimum age of 9 years, sufficient motor skills to press computer keys (the ability to tap on two adjacent keys on the computer keyboard), no major visual impairments that might interfere with performance on a computer task and comprehension of study instructions. Each participant's level of motor impairment was evaluated by trained physiotherapists using the GMFCS (Palisano et al., 1997). A detailed description of the participants is presented in Table 1.

All procedures were approved by the Ministry of Education and by the hospital's Institutional Review Board and were in compliance with ethical standards. Informed consent for participation and publication of the results was obtained by the children's parents prior to participation.

2.2. Materials and procedures

2.2.1. Raven's colored progressive matrices (RCPM)/Raven's standard progressive matrices (RSPM)

The RCPM is a measure of global cognitive performance in terms of mental age and non-verbal intelligence, addressed to children aged 5–12 years. The RCPM is considered suitable for evaluating people with severe motor impairment and speech limitations (Pueyo, Junque, Vendrell, Narberhaus, & Segarra, 2008). It is composed of 36 items divided into three sets, with each item containing a pattern with a missing piece. The child has to choose the correct missing piece out of 6–8 optional pieces, by either pointing to that piece or saying the number that represents it.

The RCPM is based on the RSPM, which is a 60-item test (divided into 5 sets) appropriate for people age 12 years and older (Raven, Raven, & Court, 2000).

Each child's *Z* score was calculated based on population data.

2.2.2. Serial reaction time task

In this task, four rectangles (4 cm × 5.4 cm) were arranged horizontally adjacent to each other on the computer screen. A red "X" changed its position from one rectangle to another according to a specific sequence about which the participants were not explicitly informed (i.e. 431241321423). Six blocks were presented with one-minute rest intervals between blocks. Each block entailed 9 repetitions of a 12 trial sequence (i.e. each block included 108 different locations of the red "X"). The first four blocks as well as the last one were *repeated sequence* blocks, while in the fifth block participants were presented with a *new sequence* (i.e. 342312143241).

Participants were asked to use one finger on their dominant hand to press the key corresponding to the rectangle in which the red "X" appeared, as quickly as possible. The use of only one finger was an adaptation of the classic SRT paradigm, intended to ease the motor overload for the CP group (Vakil et al., 2000). The stimuli presentation pace was under the participant's control. The program automatically recorded RT and errors. Two versions of the task were created to counterbalance between the *repeated sequence* and the *new sequence*, in order to rule out a specific sequence effect.

Upon completing the task, participants were questioned to assess the acquisition of declarative knowledge of the repeated sequence (i.e. "Did you notice anything while performing the task?").

Table 1
Demographic, clinical and neuropsychological information of the participants.

	Cerebral Palsy						Typically developing		
	Age	Gender	Birth weight (kg)	Gestational age (weeks)	GMFCS	Raven's progressive matrices *Z* score	Age	Gender	Raven's progressive matrices *Z* score
1	17	M	NA	NA	3	−3	17	M	2.3
2	10	F	NA	NA	4	−3	10	F	1
3	20	F	1.380	27	3	−3	18	F	1.3
4	19	F	1.900	NA	3	2	18	F	2.3
5	18	F	1.600	30	3	−3	17	F	2.7
6	10	F	0.715	29	1	−0.7	10	F	1
7	13	M	3.550	40	1	−2.3	10	M	−1
8	11	M	1.960	36	4	−3	11	M	0
9	10	M	1.355	34	1	−1	10	M	−0.7
10	16	F	2.600	38	3	−3	16	F	0.7
11	16	F	1.140	28	4	−2.3	16	F	1.7
12	16	M	1.300	29	3	−2.3	16	M	1.7
13	10	F	0.970	30	2	−3	9	F	−1.3
14	11	M	1.000	31	2	−2	11	M	0.3
15	17	F	0.915	28	1	−1.3	17	F	3
16	9	F	NA	31	3	−0.7	9	F	0
17	10	M	1.055	30	2	1.7	10	M	−0.7
18	10	M	1.255	30	3	−1.3	9	M	0.7
19	10	F	1.418	32	2	−2.3	9	F	0
20	14	F	1.440	31	2	3	14	F	3
21	12	M	0.630	27	4	0	13	M	1.7
22	10	F	1.200	28	3	−1.7	9	M	0.7
23							15	M	1

GMFCS: Gross Motor Function Classification System. M: male, F: female; NA: not available due to missing data.

2.2.3. Analysis

Three components of sequence learning in the SRT task were computed and the mean median of RT was used as the performance measure:

(1) Learning of the *repeated sequence* (the delta between score on block 1 and that of block 4).
(2) Transfer to a *new sequence* (the delta between score on block 5 and that of block 4).
(3) Recovery upon the return of the *repeated sequence* (the delta between score on block 5 and that of block 6).

A preliminary analysis was conducted to examine differences between the CP and TD groups on the Raven's IQ scores. Correlations between age, Raven's IQ scores and the three components of sequence learning in the SRT task were assessed. For the CP group, correlations were also performed between level of motor impairment and the three components of sequence learning in the SRT task. A mixed ANCOVA was conducted in order to examine the effect of group (CP, TD) and learning (blocks 1–4)/transfer to a new sequence (block 4 vs. block 5)/recovery (block 5 vs. block 6) on performance in the SRT task, with Raven's score as a covariate. The assumptions for ANCOVA were met. In particular, the homogeneity of the regression effect was evident for the covariate, and the covariate was linearly related to the dependent measure.

A chi-square analysis was conducted in order to examine independence between group (CP, Control) and declarative knowledge of the sequence (yes, no).

3. Results

While there were no differences in the age and gender of the CP and TD groups, the TD group demonstrated significantly higher non-verbal *Z* scores (Raven) ($M = 0.99$; SD = 1.24; range = −1.3 to 3) than the CP group ($M = -1.46$; SD = 1.76; range = −3 to 3), $t(43) = 5.418$, $p < 0.001$.

An overall significant correlation was found only between transfer to a *new sequence* and non-verbal IQ, $r(45) = 0.472$, $p = 0.001$, indicating that learning of the *repeated sequence* was established. With differences in intelligence between the two groups and *repeated sequence* implicit learning related to non-verbal IQ, we added the Raven test score as a covariate in all analyses of the SRT task.

3.1. Reaction time measures

Learning (*blocks* 1–4): there was an overall significant improvement in performance within the first four blocks, $F(3,126) = 7.497$, $p < 0.001$, $\eta^2 = 0.151$. In addition, there was a significant main effect for group, $F(1,42) = 5.409$, $p < 0.05$, $\eta^2 = 0.114$, indicating that the CP group ($M = 1171.24$, SD = 381.06) was significantly slower than the TD group ($M = 876.03$, SD = 379.30). Yet, learning did not interact with group $F(3,126) = 0.310$, $p > 0.05$, $\eta^2 = 0.007$, suggesting a similar learning rate in both groups (see Fig. 1, blocks 1–4).

Transfer to a new sequence (*block* 4 vs. *block* 5): no main effect was found for transfer to a new sequence, $F(1,42) = 0.537$, $p > 0.05$, $\eta^2 = 0.013$. Yet, a main effect for group reached significance, $F(1,42) = 5.292$, $p < 0.05$, $\eta^2 = 0.112$, indicating that the CP group ($M = 1125.32$, SD = 345.89) was slower than the TD group ($M = 860.28$, SD = 344.26). These findings should be interpreted cautiously due to a significant interaction between transfer to a new sequence and group, $F(1,42) = 4.164$, $p < 0.05$, $\eta^2 = 0.09$. Examination of the source of the interaction using simple analysis in each group revealed a significant increase in RT in the TD group, $F(1,21) = 13.587$, $p = 0.001$, $\eta^2 = 0.393$, but not in the CP group, $F(1,20) = 0.269$, $p > 0.05$, $\eta^2 = 0.013$ (see Fig. 1, blocks 4 and 5).

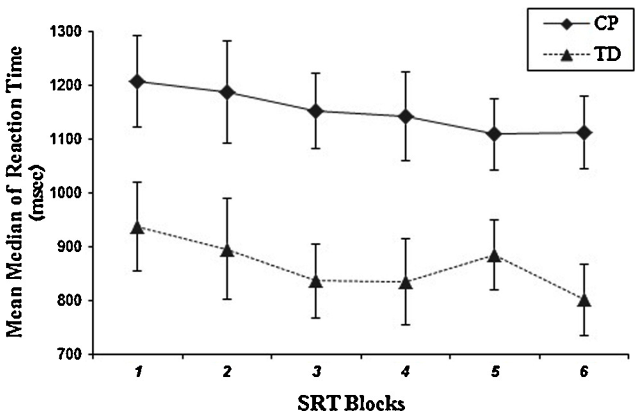

Fig. 1. Mean median of response time (RT) per block for the Cerebral Palsy (CP) and the typically developing (TD) groups. Mean median of RT and standard errors of the CP and the TD groups in the six blocks of the SRT task. Significant differences can be seen between the two groups regarding the overall RT and the response to the introduction of the new sequence in block 5.

Table 2
Partial correlations between the three components of sequence learning in the serial reaction time task.

	CP	TD
Learning & transfer	$r(19) = 0.206$, $p > 0.05$	$r(20) = 0.785$, $p < 0.001$
Transfer & recovery	$r(19) = 0.229$, $p > 0.05$	$r(20) = 0.829$, $p < 0.001$
Recovery & learning	$r(19) = 0.034$, $p > 0.05$	$r(20) = 0.678$, $p = 0.001$

CP: Cerebral Palsy; TD: typically developing.

Recovery (*block* 5 vs. *block* 6): a significant main effect for group was found, $F(1,42) = 6.727$, $p < 0.05$, $\eta^2 = 0.138$, indicating that the CP group ($M = 1110.41$, SD = 309.18) was slower than the TD group ($M = 843.34$, SD = 307.68). In addition, a significant interaction between recovery and group was found, $F(1,42) = 4.893$, $p < 0.05$, $\eta^2 = 0.104$. In examining the source of the interaction, we found that the TD group recovered following re-introduction of the repeated sequence $F(1,21) = 20.581$, $p < 0.001$, $\eta^2 = 0.495$, while there was no difference in RT between the new sequence block and the repeated sequence block for the CP group, $F(1,20) = 0.137$, $p > 0.05$, $\eta^2 = 0.007$ (see Fig. 1, blocks 5–6).

Specific learning of the *repeated sequence* can also be demonstrated by the correlations between the three components of sequence learning in the SRT task: better learning of the *repeated sequence* is accompanied by a higher "price" (i.e. increase in RT) upon replacing the sequence, and a better recovery when the original sequence is re-introduced. We therefore examined these correlations in both groups (CP, TD) (see Table 2).

3.2. Errors

Controlling for non-verbal IQ, there was no significant difference between the two groups on the total number of errors, $F(1,42) = 0.506$, $p > 0.05$, $\eta^2 = 0.012$, indicating that the CP group ($M = 22.19$, SD = 20.45) did not make more errors than the TD group ($M = 17.34$, SD = 20.35).

Results indicated that the two groups did not differ in acquiring declarative knowledge of cue-outcome associations, $\chi^2(1) = 2.670$, $p > 0.05$.

Performance on the SRT task was not age dependent, as there was no correlation between age and any of the components in either group. In addition, SRT task performance was not related to the level of motor impairment, given that none of the correlations between GMFCS and the sequence learning components were statistically significant.

4. Discussion

This study examined performance on a serial reaction time (SRT) task among children and adolescents with spastic CP compared to TD peers. We used the classical SRT task which enables the distinction between the explicit and implicit components of motor learning (Robertson, 2007). We found that although children and adolescents with CP were significantly slower than TD youth, they improved throughout the task, indicating they learned the explicit aspects. However, unlike the TD group, participants with CP did not show a decrease in RT due to the introduction of a *new sequence*, indicating that they did not implicitly learn the repeating sequence.

Our results, thus, expand those of Gagliardi et al. (2011, 2013) which purport that, impairments in implicit learning among children with CP could potentially be attributed to vulnerability in visuo-spatial processing. With only general explicit improvement in RT, and a lack of acquisition of a specific implicit sequence, our findings suggest that implicit sequence learning impairment is broader and not solely explained by a basic spatial deficiency. The use of the SRT task in our study design enables a deeper understanding of the possible source of learning impairment among children and adolescents with CP because it distinguishes between the different components of motor sequence.

Unlike earlier reports in which IQ was found not to be related to performance on the SRT task (Vicari et al., 2007), our results demonstrate that performance is associated with non-verbal intelligence. Our findings partially support previous conclusions that implicit as well as explicit learning varied with intellectual levels (Fletcher, Mayberry, & Bennett, 2000).

As in previous studies on developmental effects in sequence SL tasks (Gagliardi et al., 2013), our results confirm that performance on the SRT task is independent of age in both groups. Therefore, age was not a factor in any of our analyses.

The lack of association between severity of motor impairment, as classified by the GMFCS, and the three SL components of the SRT task, reinforces the results presented by Gagliardi et al. (2011) and those reported for an adult population with lesions to the basal ganglia (Shin, Aparicio, & Ivry, 2005). Therefore, it appears that the motor impairment per se does not explain the impairments in sequence learning, yet it highlights the common clinical knowledge that children and adolescents with CP are typically slower in performance.

Although slowness of performance is ubiquitous in children and adolescents with CP, it is not discussed in the literature. This study contributes that children and adolescents with CP perform at a much slower pace with similar learning rates to TD peers, but only with explicit learning and not implicit learning. CP participants can learn explicitly by using instructions and feedback, but they may not be sensitive to the 'hidden' sequence underlying common every-day procedures. Consequently, the order of things should be explicitly presented to them, step by step, to promote motor skill acquisition.

Gabriel, Stefaniak, Maillart, Schmitz, & Meulemans (2012) stated in a recent study that performance on an SRT task can be affected by difficulties matching the location of the stimuli on the screen to the corresponding key on the keyboard. This correspondence between the position of the stimuli on the screen and the corresponding key involves working memory (Bo, Jennett, & Seidler, 2012; Martini, Furtner, & Sachse, 2013; Vandenbossche et al., 2013) and therefore could be affected in populations with different cognitive impairments (Lejeune, Catale, Willems, & Meulemans, 2013). Individuals with CP often present difficulties in different working memory tasks (Peeters, Verhoeven, & de Moor, 2009). Thus, it could be argued that participants with CP did not learn the repeating sequence due to impairments in working memory abilities which left them invested at a more basic explicit level of task performance. Although the methodology used by Lejeune et al. (2013), which included a touch screen, might not be suitable for severely motor impaired participants with CP, further research is needed in order to assess possible influences of working memory on performance on the SRT task in this population.

5. Limitations and future directions

The current study has several limitations. While the results reported represent a relatively small sample of participants with spastic CP, the effects observed were still robust and thus should be interpreted accordingly. Additionally, in this preliminary study an imaging protocol was not conducted; hence, we relied on the diagnosis of bilateral spastic CP, considering age and severity of motor impairment, but not the specific anatomical location of the brain lesions. Clarification of possible neurological networks in CP that may impact learning abilities is required. Furthermore, combining structural and functional brain imaging with clinical evaluation of motor skill learning abilities (such as SRT) in CP is should be the goal of future studies.

Finally, to further verify the existence of a major impairment in implicit sequence learning among children and adolescents with CP, we would suggest extending the learning period (i.e. adding more blocks) and simplifying the task (i.e. three possible locations and/or a shorter sequence) in a follow up study. It is possible that participants with CP will be able to learn shorter, simpler sequences.

6. Conclusion

To conclude, our research suggests that children and adolescents with CP are impaired in implicit sequence learning; however they were able to acquire a general skill when provided with explicit instructions and sufficient practice. Since many of our basic daily activities entail a sequence of movements, and it is these precise activities that allow us to be independent in our environment, a careful examination of the manner in which children and adolescents with CP acquire such motor skills could be the basis for rehabilitation programs designed to increase their independence.

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