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Review article

A neurocognitive perspective on developmental disregard in children with hemiplegic cerebral palsy

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ABSTRACT

A common problem in children with hemiplegic cerebral palsy (CP) is the asymmetrical development of arm and hand capacity caused by the lack of use of the affected upper limb, or developmental disregard. In this paper, we provide a neuropsychological model that relates developmental disregard to attentional processes and motor learning. From this model, we hypothesize that high attentional demands associated with the use of the affected upper limb might hinder its use in daily life, and therefore may be a factor in developmental disregard. This can be assessed with a dual-task paradigm. However, until now, this has not been applied to children with CP. We provide recommendations for using a dual-task paradigm in children with CP based on empirical studies in typically developing children and children with developmental coordination disorder. Ultimately, these dual-task studies may be used to improve interventions aimed at reducing developmental disregard.

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1. Introduction

One third of the children with cerebral palsy (CP) have unilateral motor impairments, or hemiplegic CP, predominantly involving one upper and lower limb at the same side of the body. These children often use their most affected arm and hand

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less frequently and less skillfully than their least or non-affected arm and hand, which in turn limits their independence in daily life. However, the reduced capacity (i.e., the ability to execute meaningful tasks in daily life; WHO, 2001) related to the brain damage is not the only reason for the reduced spontaneous use of their affected upper limb in daily life (i.e., performance; WHO, 2001). These children also suffer from an asymmetrical development caused by the lack of use of the affected arm and hand. In children with CP, this phenomenon is referred to as "developmental disregard" (Hoare, Wasiak, Imms, & Carey, 2007). Developmental disregard (DD) can be defined as a failure to use the potential motor functions and capacities of the affected arm and hand for spontaneous use in daily life. DD is often compared to a phenomenon that may occur after a cerebrovascular accident (stroke), which is referred to as "learned nonuse" (Taub, Uswatte, & Elbert, 2002). Taub et al. (2002) explained the nonuse of the affected upper limb after stroke from a behavioral perspective, in which movements are supposed to be suppressed by negative reinforcement. In order to reverse learned nonuse, they developed a training intervention (Constraint-Induced Movement Therapy; CIMT), in which the key element is restraining the non-affected arm and hand, thereby promoting the use of the affected arm and hand through intensive training (Taub et al., 2002).

Recently, the concept of CIMT has been adjusted for children with CP and a number of review studies have been performed to examine its effectiveness (Charles & Gordon, 2005; Hoare et al., 2007; Huang, Fetters, Hale, & McBride, 2009). Although these studies found a positive trend in effectiveness, the evidence was not conclusive. One of the recommendations made in the Cochrane review by Hoare et al. (2007) was that more objective and validated outcome measures should be used to assess changes in the use of the affected upper limb in daily life (performance). In the studies reported by Hoare et al., most outcome measures used to assess performance were not accompanied by psychometric data. The only outcome measure on the functional use of the affected arm in bimanual tasks with adequate reliability, validity, and responsiveness to change was the Assisting Hand Assessment (AHA; Krumlinde-Sundholm, Holmefur, Kottorp, & Eliasson, 2007). Surprisingly, while the main aim of CIMT is to reduce DD, up to date there have been no systematic studies to determine this. Only recently, a systematic observation instrument has been developed with which DD can be assessed in a reliable and valid way (Aarts, Jongerius, Geerdink, & Geurts, 2009) following modified CIMT (mCIMT) combined with bimanual training in children with hemiplegic CP (Aarts, Jongerius, Geerdink, van Limbeek, & Geurts, 2011). In this study, the training intensity of original CIMT (i.e., minimally 3 h of therapy a day for 2 weeks) was adjusted to be suitable for children (3 h of therapy a day, 3 days a week for 6 weeks). Aarts et al. (2011) found that only children with poor manual ability (score III on the Manual Ability Classification System; Eliasson et al., 2006) improved significantly on the DD score, whereas children with better manual ability hardly showed any changes on this measure. In fact, even though some of these children showed improvements in capacity, they still underused their affected arm and hand. This raises the question as to what underlying processes, besides negative behavioral reinforcement, are involved in DD. In this paper, we provide an explanation using a neuropsychological model that relates motor learning to cognition. The crucial elements of this model are the attentional processes associated with motor learning.

2. Relationship between motor learning and cognition

Rehabilitation may be regarded as the (re)learning of motor control (Krakauer, 2006). In children with CP it is not as much relearning (as is the case after stroke), but rather learning of movements of the affected arm that have never, or hardly ever, been performed before. Fitts and Posner proposed a theory of motor learning that has been widely accepted (Fitts & Posner, 1967). Their theory proposes three phases of motor learning; the cognitive, associative, and automatic phase. A central tenet of this theory is that improvements in a motor skill run parallel with a decrease in cognitive or attentional resources needed

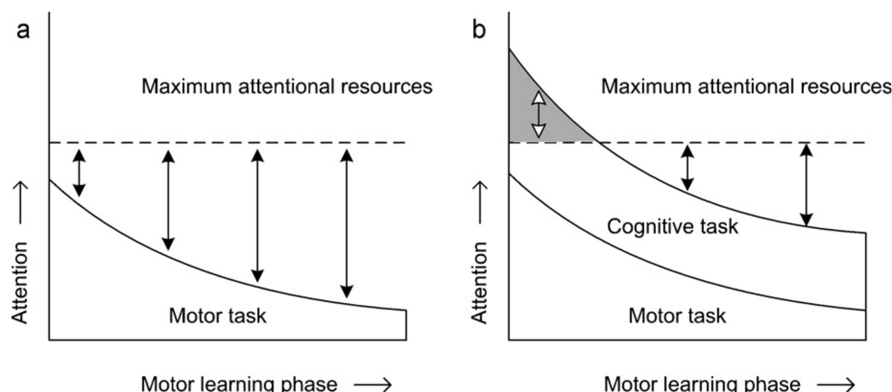

Fig. 1. Theoretical model on the relationship between motor learning and attentional resources. In the early phase of motor learning, a large part of the available attentional resources are needed to perform the motor task (panel a). During the final phases of motor learning, attention gradually decreases until the motor task is performed almost automatically. If the motor task is sufficiently automated and adequate attentional resources remain available (indicated by the black arrows), then performing a cognitive task concurrently with a motor task does not interfere with the motor task (panel b). If the motor task, however, is not sufficiently automated, dual-task interference occurs (indicated by the white arrow in panel b), over-demanding the attentional resources and resulting in errors or delays in the performance of either or both tasks.

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to perform the task (Fig. 1a). In the third and final phase, the motor skill requires only a minimal amount of attention and the skill is said to be automated (Wickens, 2008).

This close relationship between cognitive processes and motor-skill learning has important implications for functional recovery (Mulder, Zijlstra, & Geurts, 2002). It was empirically shown that patients who have to (re)learn a lost motor skill following acquired brain damage such as stroke, or lower-limb amputation, needed a high level of attention in the early stages of rehabilitation (Cockburn, Haggard, Cock, & Fordham, 2003; Geurts & Mulder, 1994). Only when the attentional load was reduced and the skill became increasingly automated, functional recovery was complete and the skill could be used with little mental effort in daily life. We argue that in children with CP the discrepancy between capacity and performance of the affected upper limb in daily life, i.e. DD, coincides with an excess in cognitive effort that is associated with motor control of the affected upper limb. Hence, in our explanation, we explicitly link behavior (motor learning) with brain processes (attention). That is, if a task performed with the affected upper limb is not automated and therefore requires a disproportional amount of attentional resources, it is likely to be avoided in daily life, and consequently performed with the least or non-affected side. It is exactly this aspect of performance that CIMT tries to overcome.

3. Dual-task paradigm

A well-established paradigm with which the level of automaticity of a motor task can be assessed is the dual-task paradigm. Here, a motor task is executed while at the same time an attention-demanding cognitive task needs to be performed (Kahneman, 1973). According to Kahneman, attention is limited and if the motor task requires too much of the maximally available attentional capacity, errors or delays in the performance of one or both tasks are observed. This is referred to as dual-task interference (Kahneman, 1973). The amount of interference depends on the level of attention that is needed for the motor and the cognitive task. If a motor task is adequately learned, few attentional resources are needed to perform the task, thereby leaving sufficient resources for the performance of concurrent attention-demanding tasks. However, if a motor task is not sufficiently automated and requires a disproportional amount of attentional resources, the performance of both tasks cannot be maintained and dual-task interference occurs (Fig. 1b). Dual-task paradigms have been extensively used in rehabilitation research in patients with acquired brain damage (e.g. stroke), patients with Parkinson's disease, balance-impaired elderly, and healthy younger and older adults (Woollacott & Shumway-Cook, 2002). In general, a dual-task paradigm is viewed as a more accurate representation of daily life than a controlled laboratory setting without any concurrent cognitive task. It may therefore provide a more sensitive assessment of functional recovery. For instance, in a study on recovery of standing balance after stroke, the effects of a concurrent arithmetic task were tested on, among other factors, the ability to load the affected leg (de Haart, Geurts, Huidekoper, Fasotti, & van Limbeek, 2004). It was found that, although weight-bearing asymmetry significantly improved after training in the single-task condition, it remained consistently higher in the dual-task condition over the course of rehabilitation. In other words, patients may have consciously learned to distribute their weight more symmetrically, but this was not fully automated. During a concurrent attention-demanding task the patients tended to rely more on their non-affected leg. These results underscore the importance of measuring automaticity of motor control via dual-task conditions, which would remain undetected under single-task conditions.

To evaluate the efficacy of CIMT in children with CP, it is also of eminent importance to assess the level of automaticity of motor control. While the majority of studies using (m)CIMT showed improvements of capacity of the affected upper limb after the intervention (Charles & Gordon, 2005; Hoare et al., 2007; Huang et al., 2009), there is still a void in research scrutinizing the long-term effects on actual use of the affected side in daily life. The next generation intervention programs need to consider the attentional efforts associated with the motor tasks in such a way that better insights can be obtained in the underlying factor(s) of DD. Starting from our neuropsychological model we argue that the origin of DD may, at least in part, be sought in the attentional resources associated with the motor performance, even after weeks of intensive rehabilitation.

3.1. Dual-task paradigms in children with CP

In order to provide an overview of the research that used dual-task paradigms for children with CP, we conducted a systematic search in the Pubmed, Medline, PsychINFO and Embase databases using the following key words: (1) dual task, or concurrent task; combined with (2) cerebral palsy, or congenital. Studies were qualified as dual-task studies when a motor task was combined with a cognitive task. We found only one study that used a dual-task paradigm in children with CP. This study was on postural control and not on upper-limb control (Reilly, Woollacott, van Donkelaar, & Saavedra, 2008b). In this study, children with bilateral CP (10–14 years old) were compared to a group of typically developing older children (7–12 years old) and typically developing younger children (4–6 years old). All children performed two postural control tasks (wide and narrow stance), with a concurrent cognitive task scaled to the individual ability (visual working memory task). Results indicated that children with CP, like the younger children, were more unstable in narrow stance and performed worse on the memory task than the older children. Although postural control in the wide stance did not differ between children with CP and older children, children with CP, like the younger children, demonstrated more dual-task interference when the cognitive task was added compared to the older children. Thus, the dual task was a sensitive measure of automaticity of postural control.

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Obviously, this literature search lays bare a void in studies on motor control and attentional load in CP. From our reasoning above it is evident that advances need to be made in the research area of upper-limb motor control and attentional load in CP. Therefore, in the remainder of this article, we will provide recommendations for the use of a dual-task paradigm in children with CP. First, we will discuss the use of dual-task experiments in typically developing children, to assess the general ability of young children to perform multiple tasks. Second, we will discuss dual-task experiments in children with a congenital motor disorder that is sometimes referred to as "mild CP": developmental coordination disorder (DCD). Third, by combining these findings we provide recommendations for future research in children with CP.

3.2. Dual-task paradigms in typically developing children

A prerequisite for performing dual tasks is the ability to divide attention between the two tasks. Dividing attention is a function of 'executive attention', which entails processes involved in the regulation of thought, emotion and behavior (Posner & Rothbart, 1998). These control processes improve exponentially with age, with most changes occurring between 8 and 12 years of age (Kail, 1991). The ability to focus attention on a task goal while ignoring irrelevant sources of information improves most markedly between 6 and 10 years of age (Strutt, Anderson, & Well, 1975; Mezzacappa, 2004). Still, children as young as 3 years old were able to perform conflict resolution in a modified Stroop paradigm, which is also a function of executive attention (Posner & Rothbart, 1998). In the original Stroop task, subjects are instructed to name the color of the ink of a presented word, which can be congruent (i.e., the word 'red' in the color red), incongruent (i.e., the word 'red' in the color green), or neutral (i.e., the word 'table' in the color red). These studies indicate that children as young as 3 years old were able to divide attention and resolve conflicts. Nevertheless, these tasks should be tuned to the child's individual level, as younger children have a smaller overall capacity of the executive attentional system (Irwin-Chase & Burns, 2000; Reilly, van Donkelaar, Saavedra, & Woollacott, 2008a). In order to determine dual-task interference (i.e., performance on the dual task relative to the single task), the single-task performance has to reach a certain level of accuracy to allow for possible changes in performance during the dual task compared to the single task (Rueda et al., 2004).

Recent dual-task studies with typically developing children showed that children from 4 to 12 years old were capable of performing dual tasks during postural control (Blanchard et al., 2005; Olivier, Cuisinier, Vaugoyeau, Nougier, & Assaiante, 2007; Reilly et al., 2008a; Schmid, Conforto, Lopez, & D'Alessio, 2007; Schaefer, Krampe, Lindenberger, & Baltes, 2008) and walking (Cherng, Liang, Hwang, & Chen, 2007; Huang, Mercer, & Thorpe, 2003). All studies showed dual-task effects. Still, protocols and outcome variables varied among the different studies. In the cognitive tasks, for instance, the repeating of numbers or words, counting, remembering words or shapes, and conflict resolution in a modified Stroop task were used. Two studies found increases in postural sway during the dual-task condition in children between 8 and 10 years of age, but did not report the effects on the cognitive task (Blanchard et al., 2005; Schmid et al., 2007). Other studies compared children with healthy young adults, and used motor and cognitive tasks with different levels of difficulty (Olivier et al., 2007; Reilly et al., 2008a; Schaefer et al., 2008). In general, they found a lower overall motor and cognitive performance in the children (7–12 years old), even more so in younger children (4–6 years old) and with increased task difficulty (Cherng et al., 2007; Huang et al., 2003).

3.3. Dual-task paradigms in children with DCD

There have been some studies using a dual-task paradigm in children with DCD with results similar to those of typically developing children. Studies on postural control found more dual-task interference in children with DCD than in typically developing children (Laufer, Ashkenazi, & Josman, 2008; Tsai, Pan, Cherng, & Wu, 2009). One study on walking did not report such a difference (Cherng, Liang, Chen, & Chen, 2009). During a manual tracing task in which children with DCD, children with ADHD, and typically developing children had to draw a straight line within a 2 mm wide path, no dual-task interference was found (Miyahara, Piek, & Barrett, 2006). Although different cognitive tasks were used that were expected to result in different levels of dual-task interference (i.e., distraction using background music and a telephone ring sound, counting upwards in steps of one, and naming animals that were presented next to the drawing path), no performance decline on the tracing task was observed in any of the groups. These results suggest that the cognitive tasks were not enough attentionally demanding to result in dual-task interference.

4. Main points of focus in a dual-task paradigm for children with CP

In sum, previous studies showed that typically developing children and children with DCD were able to perform dual tasks, and that dual-task interference was generally higher in children with DCD. Therefore, application of this methodology in children with CP demands careful selection of the cognitive as well as the motor task, as not all combinations will lead to interference. In children with DCD, several cognitive tasks resulted in the expected dual-task interference during postural control. These were: naming visually presented objects, counting backwards in steps of three, naming the pitch of a tone (high or low), and remembering food items. However, during walking, repeating series of digits forwards and backwards resulted in equal levels of dual-task interference in children with DCD and typically developing children, indicating that this task was too difficult for both groups. During a manual task, however, no dual-task interference effects were found in children with DCD, children with ADHD, and typically developing children. The cognitive tasks used in this manual tracing

experiment (distraction, counting forwards with steps of one, naming animals) may have been too easy in combination with the manual task. Another key factor may be that, obviously, the consequences of failure to perform the manual task are not as detrimental as during walking and balance control. For children with CP, these results have to be taken into consideration when designing an upper-limb dual-task paradigm. The level of difficulty of the cognitive task should possibly be more difficult than merely distraction, counting and naming objects, but not as difficult as remembering series of digits. In addition, children with CP often have problems in executive functions, such as sustained and divided attention (Bottcher, Flachs, & Uldall, 2010) and inhibitory control (Christ, White, Brunstrom, & Abrams, 2003). Thus, the cognitive task should be scaled to the level of the individual child. Finally, a visual working memory task was found to be suitable for children with CP in a postural control task (after scaling the difficulty to the individual child), but together with a manual task, a visual task may yield different results. A manual task relies heavily on visual control and adding a concurrent visual cognitive task may result in structural interference instead of generalized attention capacity interference. Structural interference takes place if two tasks are controlled by similar specific sensory or motor subsystems, whereas generalized attention capacity interference takes place when two tasks rely on the same attentional resources (Kahneman, 1973). Clearly, finding the best combination of a motor and a cognitive task in a dual-task paradigm for a specific patient group of a certain age is an empirical question of great challenge that requires taking into account existing empirical evidence and theoretical considerations on the relation between brain and behavior.

In addition to the choice of tasks, it is important to search for the most sensitive outcome measures in the tasks that reflect interference effects. Most upper-limb motor tasks have temporal and spatial accuracy demands that show a speed-accuracy trade-off (Schmidt & Lee, 2005). The performance of many cognitive tasks may show similar speed-accuracy trade-off effects, for instance between speed and errors. In addition to these within-tasks trade-off effects, there are the between-tasks trade-off effects inherent in all dual-task paradigms. This complexity of outcomes warrants a thorough (a priori) consideration of the use of composite scores that integrate within-tasks trade-off effects as well as of statistical approaches to use such composite scores into one overall analysis.

In this respect, instructions given to the participants are essential. Should they value one task over the other in the case of difficulty, or is the execution of both tasks equally important? Is accuracy more important than speed within a task or vice versa? Only with proper instructions, a fair interpretation of the results is possible, both on an individual and on a group level.

5. Implications for future research

Many rehabilitation programs for children with hemiplegic CP aim at reducing DD. However, while there is substantial evidence for positive effects of e.g. (m)CIMT on upper-limb motor *capacity*, there is not yet convincing evidence that such rehabilitation programs are effective in decreasing DD. In this paper, we propose a neuropsychological model from which we hypothesized that attentional demands associated with the use of the affected upper limb might hinder its use in daily life, despite improvements in capacity after intensive training. A dual-task paradigm consisting of an upper-limb motor task combined with a cognitive task can be used in future studies as a means to assess automaticity of motor control following an intervention. In addition, neuroscientific methods such as electroencephalography and event-related potential registration may be applied to scrutinize and directly measure the cognitive effort during motor performance (Rugg & Coles, 1995).

Finding an answer to the question whether lack of automaticity plays a role in DD is a necessary next step in improving long-term efficacy of rehabilitation in children with hemiplegic CP. Indeed, the ultimate goal of upper-limb rehabilitation is the reduction of DD and the improvement of spontaneous use of the affected limb in daily life. Therefore, dual-task paradigms should be implemented in intervention programs, so that the cognitive effort associated with upper-limb control can be closely monitored and rehabilitation programs can become fine-tuned to the individual child. One training aspect that may be improved is the intensity of practice, such as frequency and duration. While basic capacity may be optimized within a short training period, automatization of this capacity may take much longer, i.e., follow a different time scale of learning, and thus require a different type of training. In current CIMT programs shaping is an important aspect to improve capacity. Still, the reduction of DD may in fact benefit more from practice under various complex (dual-task) conditions (Mulder & Hochstenbach, 2001; Schmidt & Lee, 2005). For instance in sports, substantial repetitive training is needed for a novice to achieve a certain skill level. However, a great number of matches and tournaments that are played in various circumstances are required to develop a truly robust level of skilled performance that can be maintained even under stressful or distracting circumstances. Upper-limb training should, therefore, not end after an intensive rehabilitation program, but be continued and integrated in the daily-life activities of children with CP.

6. Conclusion

Based on a neuropsychological model and existing empirical evidence in individuals with acquired brain damage, we hypothesize that lack of automatization of movement might be a crucial underlying factor for DD in children with hemiplegic CP, even after intensive rehabilitation. However, this has not yet been substantiated by empirical studies in this group. Intensive training programs that use complex (dual-task) conditions should be investigated for their efficacy in this respect. Dual-task studies with typically developing children and children with DCD indicate that dual tasking is applicable in young children with and without motor disorders. Therefore, dual-task assessment should be considered as an important outcome

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measure to investigate automaticity of upper-limb motor control in children with CP before and after training interventions. As it cannot be expected that a standardized dual-task protocol will become available for children of different age and severity, well considered motor and cognitive tasks must be developed, together with valid outcome measures. These can build on studies conducted in typically developing children and children with DCD. Furthermore, parallel to investigating automaticity of upper-limb motor control, advancements need to be made in improving automaticity during intervention programs with the ultimate goal of improving spontaneous use of the affected upper limb in daily life.

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