Impaired motor planning and motor imagery in children with unilateral spastic cerebral palsy: challenges for the future of pediatric rehabilitation

BERT STEENBERGEN1 | MARJOLEIN JONGBLOED-PEREBOOM1 | STEFFIE SPRUIJT1 | ANDREW M GORDON2

1 Behavioural Science Institute, Radboud University Nijmegen, Nijmegen, the Netherlands. 2 Department of Biobehavioral Sciences, Teachers College, Columbia University, New York, NY, USA.

Correspondence to Dr Bert Steenbergen, Behavioural Science Institute, Radboud University Nijmegen, Nijmegen, the Netherlands. E-mail: b.steenbergen@pwo.ru.nl

Motor impairments of the affected side in unilateral spastic cerebral palsy (USCP) are an important cause of activity limitations. Therefore, an obvious goal of pediatric rehabilitation in young children with USCP is to train the affected side of the body so as to facilitate its use to perform the manifold of daily tasks. In most such rehabilitation programs, such as constraint-induced movement therapy, the affected side is intensively trained for an extended period, sometimes totaling 60 hours or more. These programs are based on the principle of neural plasticity: sensorimotor deprivation of a limb leads to a decreased cortical representation (the ‘use it or lose it’ phenomenon), whereas sensorimotor stimulation leads to a use-dependent cortical reorganization. In general, these intensive training programs have been effective in improving the capacity of the affected body side. Although most rehabilitation programs for children with USCP primarily focus on improving the capacity of the affected side, more recent programs have been developed in which bimanual activities are also trained to improve functional use of the affected side. One such example is the hand-arm bimanual intensive training (HABIT) program, in which 2 to 3 weeks of bimanual training is provided.

Although these concerted rehabilitation efforts improve capacity and functional use of the affected side, recent evidence suggests that motor planning deficits are also a possible underlying cause for compromised performance of activities of daily living. In this paper we will first review the growing evidence for the motor planning deficit, followed by studies on motor imagery in this population. Motor imagery is an experimental approach in which the contents of the motor plan become overt. Converging evidence indicates a compromised motor imagery ability in USCP. As the neural structures of both motor planning and motor imagery overlap, rehabilitation by motor imagery training may alleviate motor problems in USCP. Increasing evidence for this approach exists in older adults with stroke. We conclude this review with recommendations on such a training approach for children with USCP.

Compromised action performance is one of the most characteristic features of children with unilateral spastic cerebral palsy (USCP). Current rehabilitation efforts predominantly aim to improve the capacity and performance of the affected arm. Recent evidence, however, suggests that compromised motor planning may also negatively affect performance of activities of daily living. In this paper we will first discuss the recent evidence for this motor planning deficit, followed by studies on motor imagery in this population. Motor imagery is an experimental approach in which the contents of the motor plan become overt. Converging evidence indicates a compromised motor imagery ability in USCP. As the neural structures of both motor planning and motor imagery overlap, rehabilitation by motor imagery training may alleviate motor problems in USCP. Increasing evidence for this approach exists in older adults with stroke. We conclude this review with recommendations on such a training approach for children with USCP.

MOTOR PLANNING DEFICITS IN USCP

Motor planning is the ability to anticipate the end of the upcoming action when preparing a movement towards an object. Theoretically, motor planning, or action prediction, is associated with internal forward modeling. An internal model is a neural system that simulates the upcoming action. Thus, before the actual motor performance, a prediction of the action and its sensory consequences is made based on the efference copy of motor commands. This allows the central nervous system to maintain stability as it predicts and corrects movements before afferent signals are received. Consequently, impairments in forward modeling may result in a movement system that is more dependent on the slow afferent system, rather than the predictive efferent copy. Such an impairment in forward modeling (termed the ‘internal modeling deficit’) has increasingly been advocated as a cause for performance deficits that children with developmental coordination disorder encounter.

A frequently used experimental set-up to study motor planning purports picking up an object with the intention of placing it at a specific goal position and/or in a specific orientation. In general, participants sacrifice the postural comfort of the initial grip with which the object is picked up, such that the task is completed with the hand and arm in a comfortable posture (Fig. 1). The need for efficient motor planning becomes even more stringent in sequential tasks, where an object is manipulated to use it as a tool. Obviously, most tasks in daily life have a sequential component. Accumulating evidence indicates that individuals with USCP have compromised motor planning abilities, even when performing tasks with their less affected side. In one of the first stud-
ies on motor planning in USCP, Mutsaarts et al.\textsuperscript{16} used a six-sided object that participants had to grasp and orient to match a pre-instructed orientation, by rotating it either 60°, 120°, or 180°. Especially in the 180° rotation condition, participants had to anticipate the end of the action to perform this task correctly, because failing to adjust the grip orientation with which the object was grasped led to a failure in performing the task (Fig. 2). The results showed that adolescents with USCP failed to perform the task with their less-affected hand correctly, owing to an impaired motor planning process. This finding has since been replicated in several follow-up studies using different set-ups.\textsuperscript{17} An additional and consistent finding is that planning problems are more severe when the right body side is affected, thus when brain damage is probably most dominant in the left hemisphere.\textsuperscript{7} These findings are in line with neuroimaging studies in healthy adults that show a left hemisphere dominance for action planning.\textsuperscript{18}

**MOTOR IMAGERY IN USCP**

To examine whether a deficit in internal models may cause the compromised motor planning, motor imagery can be used.\textsuperscript{15} According to Jeannerod,\textsuperscript{19} motor imagery is closely related to the motor representations involved in the planning and execution of movements. By definition, motor imagery is the internal rehearsal of an upcoming motor action within working memory without any overt motor output.\textsuperscript{20} Thus, the actual execution of the motor action is inhibited. Motor imagery can thus serve as an experimental tool to reveal the contents of the internal model. An often-used manner of assessing motor imagery capacity is the hand laterality task. In this task, a hand is shown in different orientations to the participant. The participant is asked to decide as quickly as possible whether the displayed hand is a left or a right hand (see Fig. 3 for examples of stimuli in a hand laterality task). Typically, reaction times increase with an increase in rotation angle of the displayed hand. This result indicates that the participant performed the task by mentally rotating the stimulus. However, to be sure that the participant is indeed involved in motor imagery, namely the mental rotation of the object in the remembered representation of the movement, a task that requires the use of working memory, additional methods have to be used.

**What this paper adds**

- An overview of recent research on motor planning and motor imagery in CP.
- Recommendations for the application of motor imagery training in children with CP.
of his/her own hand, additional evidence is required. If motor imagery is used, the reaction time profile should be subject to the same biomechanical constraints as actual movements. For example, hand rotations in the medial direction (towards the mid-sagittal plane) are biomechanically easier to perform than hand rotations in the lateral direction. If participants use motor imagery in the hand laterality task, this difference between medial and lateral rotations should also be evident in the reaction time profile. This was indeed shown to be the case in recent studies with healthy adults.21 Similarly, it has been shown that one’s posture has a specific effect on the performance on the hand laterality judgment task.22

Studies on motor imagery in USCP are not widespread, but the evidence thus far suggests that this ability is compromised in individuals with CP.23,24 For example, in a recent study, Crajé et al.25 failed to find lateral–medial rotation effects in participants with USCP despite the fact that these participants were able to perform the task above chance level. This finding suggests that their ability to use motor imagery to solve the task is compromised.

REHABILITATION BY MOTOR IMAGERY TRAINING IN USCP?

The combined findings on motor planning and motor imagery deficits in USCP begs the question as to whether these two processes are causally related. If so, then training of motor imagery may positively affect the internal model and in turn could improve motor performance. The possible causal relation between both processes can be delineated from two lines of evidence. First, there is a considerable neural overlap between both processes. Second, the efficacy of motor imagery training has been repeatedly shown in adults with stroke.

First, with respect to the neural overlap, neuroimaging studies provide evidence for activation of similar neural structures during motor imagery and the actual performance of the same movement. Specifically, activation in the primary motor cortex (M1), premotor cortex, supplementary motor areas, and parietal lobe was found during motor imagery and actual movement (for recent meta analysis see Zacks26). Moreover, the role of the parietal cortex was evident from patients with lesions in this structure. These patients showed impaired performance on a task that demanded imagery of a manual motor task.27 These collective findings indicate that motor imagery and motor planning may be subserved by the same neural structures.

Second, beneficial effects of motor imagery training for rehabilitation of upper limb function have been reported in adult patients with stroke (for reviews see Dickstein and Deutsch28 and Nilsen et al.29). In general, training sessions entail that participants are instructed to imagine movements with the affected arm. Retention of the positive effects on upper limb functioning was shown even after a 3 month retention interval.10,21 These findings from adult stroke rehabilitation suggest that the relation between motor imagery and motor performance may be causal in nature. Finally, in a study with children with developmental coordination disorder, beneficial effects of motor imagery training were shown on motor performance.10 Given the efficacy of motor imagery training for upper limb rehabilitation in adult patients with stroke and children with developmental coordination disorder, such training may be a useful addition to rehabilitation programs for children with USCP.31 Despite its theoretical feasibility, motor imagery training still awaits empirical testing in USCP.

IMPLICATIONS FOR PEDIATRIC REHABILITATION

The first issue that needs to be resolved before using motor imagery training in children with USCP is the age at which young children are able to use motor imagery. Several studies have been performed in typically developing children (between 5 and 12 y old) to assess their ability to use motor imagery.32–34 Owing to differences in methodology and interpretation of results, definite conclusions about the age at which children are able to use motor imagery are not warranted. It is clear, however, that the age range between 5 and 10 years is critical for the development of motor imagery.

A second issue is the set-up with which motor imagery is tested and the way in which it is trained: that is, the specific task context. To test motor imagery capacity, most of the studies in children make use of the hand laterality task or the mental chronometry task. In the latter, the durations of the actual performance and the imagined performance are compared with high correlations implying the use of motor imagery. Still, for young children such a context may in fact cloud their real capacity. Therefore, experimental set-ups to test and train motor imagery in adults are probably not suitable for children. More specifically, the study and possible training of motor imagery should proceed in a context that is meaningful to the child and relates to their developmental age. A case in point here is observational learning. It is beyond the scope of this paper to discuss in detail the possible benefits of observational learning, such as mirror box therapy.35 However, it is clear that the discovery of the mirror neuron system in humans may have implications for therapy. Specifically, observation of others performing movements may facilitate rehabilitation of one’s own motor performance. In this respect, action observation therapy was recently proposed as a possible effective therapy for children with USCP.36

A third issue that is important before applying motor imagery training as an additional technique in pediatric rehabilitation of children with CP is dosing of training. The use and training of the affected side may be associated with high attentional demands, as was shown in patients with stroke using their affected side in a dual-task.18 As the attention span of young children is limited, multiple short sessions may be more effective than fewer long ones. Additionally, because children with CP often have working memory problems39 the benefit of short sessions is even more warranted.

A fourth issue is the type of motor imagery training. Because of the aforementioned working memory problems, the child should not be overloaded with instructions, as is the case in adults, rather, the training should be done in a more implicit manner. For example, a small cartoon may demonstrate and guide the child through the imagery process. Finally, it has to be established to what extent the capacity to perform motor imagery is dependent upon IQ, which is one of the known comorbidities of CP. Currently it is not known whether IQ affects motor imagery capacity in children.

CONCLUSION

Motor imagery training may be a valid and useful tool to complement upper limb rehabilitation in young children with CP. Although it has been shown to be beneficial in adult patients with stroke and it is theoretically feasible to train the internal model, it still awaits empirical testing in young children with CP. Some important individual aspects that need to be taken into account for setting up such a training protocol are age, attention, working...
Importantly, recent studies have shown that motor planning deficits are amenable to training effects,\(^{40}\) suggesting promise for this approach.

**CONFLICTS OF INTEREST**

The authors report no conflicts of interest with this paper.