UNIVERSIDADE FEDERAL DE MINAS GERAIS
INSTITUTO DE CIÊNCIAS BIOLÓGICAS
PROGRAMA DE PÓS-GRADUAÇÃO EM NEUROCIÊNCIAS

Deisiane Oliveira Souto

NEUROMOTOR IMPAIRMENTS IN CHILDREN WITH HEMIPLEGIC CEREBRAL PALSY: EVALUATION, MECHANISMS AND INTERVENTION.

Alterações neuromotoras em crianças com paralisia cerebral hemiplégica: avaliação, mecanismos e intervenção.

BELO HORIZONTE - MG
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NEUROMOTOR IMPAIRMENTS IN CHILDREN WITH HEMIPLEGIC CEREBRAL PALSY: EVALUATION, MECHANISMS AND INTERVENTION.

DEISIANE OLIVEIRA SOUTO

Tese submetida à Banca Examinadora designada pelo Colegiado do Programa de Pós-Graduação em NEUROCIÊNCIAS, como requisito para obtenção do grau de Doutor em NEUROCIÊNCIAS, área de concentração NEUROCIÊNCIAS BÁSICAS.

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RESUMO

A paralisia cerebral (PC) é causada por um dano multifatorial não progressivo que afeta o sistema nervoso central em desenvolvimento. O subtipo espástica é o mais comum e resulta de lesões no sistema piramidal. Embora a PC Hemiplégica (PCH) cause danos ao sistema piramidal unilateral, a apresentação clínica de seus déficits neuromotores também é sugestiva de sintomas extrapiramidais e cerebelares bilateralmente. É possível que os comprometimentos motores bilaterais observados em crianças com PCH estejam relacionados a déficits no planejamento motor. Estudos de neuroimagem mostraram que imagem motora (IM), ou seja, movimentos imaginados, ativa redes neurais semelhantes às envolvidas no planejamento e na execução dos movimentos, podendo ser uma ferramenta potencial na reabilitação dos déficits neuromotores observados na PCH. Embora IM tenham sido apontada como uma estratégia promissora para a reabilitação, ainda são escassas as informações sobre seu desenvolvimento ao longo da infância e adolescência. Não está claro ainda se mesmo após lesão cerebral precoce as crianças com PCH podem se envolver em tarefa que exige o uso de IM. O objetivo geral da presente dissertação é fazer uma investigação ampla dos comprometimentos neuromotores na PCH com ênfase no estudo da IM e sua efetividade na reabilitação dos membros superiores dessa população. Com este propósito, o presente trabalho é composto por quatro estudos. No Estudo 1 foi estabelecido um protocolo de exame neuromotor capaz de avaliar a presença de comprometimentos bilaterais nos três níveis de integração motora (piramidal, extrapiramidal e cerebelar) de crianças com PCH. Nesse estudo, as crianças com PCH apresentam sinais neuromotores sugestivos de comprometimentos nos três níveis de integração do sistema motor. Além disso, os comprometimentos foram reportados em ambos os membros, independente da lateralidade da lesão. No Estudo 2, foi utilizada uma tarefa de rotação mental para avaliar o desenvolvimento da IM em crianças e adolescentes saudáveis de diferentes faixas etárias (6-7 anos, 8-9 anos, 10-11 anos, 12-13 anos) comparando seu desempenho ao de adultos saudáveis. Os resultados mostraram que as crianças de 6 a 7 anos já são capazes de realizar IM, contudo, ocorre uma melhora progressiva conforme o aumento da idade. Foi descoberto ainda que aos 10 anos de idade a habilidade de IM das crianças é semelhante aos adultos. O Estudo 3 investigou a capacidade de IM em crianças e adolescentes com PCH utilizando uma tarefa de rotação mental. Foi descoberto que as crianças com PCH podem desempenhar IM, no entanto, sua habilidade não equivale a de controles saudáveis. Além disso, foi verificado que a habilidade de IM pode ser influenciada pelo desempenho funcional e pela memória de trabalho das crianças. Por fim, o Estudo 4 avaliou a eficácia de um protocolo de reabilitação baseado no treinamento de IM associado a prática física na reabilitação dos membros superiores de crianças com PCH. Diferenças significativas foram encontradas entre o grupo intervenção e controle, fornecendo evidências preliminares da eficácia de IM na reabilitação neuromotora. Esses resultados serão discutidos e interpretados considerando a literatura atual e suas implicações para a prática clínica e a neuro-reabilitação.

Palavras-chave: paralisia cerebral hemiplégica, comprometimentos, exame neuromotor, imagética motora, membro superior, reabilitação.

ABSTRACT

Cerebral palsy (CP) is caused by non-progressive multifactorial damage that affects the developing central nervous system. The spastic subtype is the most common and results from lesions to the pyramidal system. Although hemiplegic CP (HCP) causes damage to the unilateral pyramidal system, the clinical presentation of neuromotor deficits also suggests bilateral extrapyramidal and cerebellar symptoms. It is possible that bilateral motor deficits observed in children with HCP are related to motor planning deficits. Neuroimaging studies have shown that motor imaging (MI), i.e., imagined movements, activates neural networks similar to those involved in planning and executing movements, and can be a potential tool in the rehabilitation of neuromotor deficits observed in HCP. Although MI has been identified as a promising strategy for rehabilitation, information on its development during childhood and adolescence is still scarce. It is still unclear whether, even after brain damage, children with HCP can be involved in a task that requires the use of MI. The general objective of this dissertation is to make a broad investigation of neuromotor deficiencies in HCP emphasizing the study of MI and its effectiveness in the rehabilitation of the upper limbs of this population. For this purpose, the present work consists of four studies. In Study 1, a neuromotor examination protocol was established, capable of assessing the presence of bilateral deficiencies in the three levels of motor integration (pyramidal, extrapyramidal and cerebellar) of children with HCP. In this study, children with HCP present neuromotor signs suggesting deficiencies in the three levels of motor system integration. In addition, deficiencies were reported in both limbs, regardless of the laterality of the injury. In Study 2, a mental rotation task was used to evaluate the development of MI in healthy children and adolescents of different age groups (6-7 years, 8-9 years, 10-11 years, 12-13 years) by comparing their performance in healthy adults. The results showed that children 6-7 years of age are already able to perform MI, but there is progressive improvement as age increases. It was also detected that at age 10, children's MI skills are similar to those of adults. Study 3 investigated MI capacity in children and adolescents with HCP using a mental rotation task. It was found that children with HCP can play MI, however, their ability does not match that of healthy controls. Furthermore, it was found that MI capacity can be influenced by children's functional performance and working memory. Finally, Study 4 assessed the effectiveness of a rehabilitation protocol based on MI training associated with physical practice in the rehabilitation of upper limbs of children with HCP. Significant differences were found between intervention and control groups, providing preliminary evidence of the efficacy of MI in neuromotor rehabilitation. These results will be discussed and interpreted considering the current literature and its implications for clinical practice and neuro-rehabilitation.

Keywords: hemiplegic cerebral palsy, impairments, neuromotor examination, motor imagery, upper limb, reabilitation.

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LIST OF ABBREVIATIONS AND ACRONYMS

CP	Cerebral Palsy
HCP	Hemiplegic Cerebral Palsy
CNS	Central Nervous System
MI	Motor Imagery
MACS	Manual Ability Classification System
PANESS	Physical and Neurological Examination for Soft Signs
ZNA	Zurich Neuromotor Assessment
RHCP	Right hemiplegic cerebral palsy
LHCP	Left hemiplegic cerebral palsy
UCP	Unilateral Cerebral Palsy
RUCP	Right Unilateral Cerebral Palsy
LUCP	Left Unilateral Cerebral Palsy
SPSS	Statistical Package for Social Sciences
9-HPT	Nine Hole Peg Test
Raven's CPM	Raven's Coloured Progressive Matrices
IQ	Intelligence Coefficient
HLJ	Hand Laterality Judgment
RT	Reaction Time
RTs	Reaction times
DCD	Development Coordination Disorder
M	Mean
SD	Standard Deviation
WISC IV	Wechsler Intelligence Scale for Children fourth Edition
AHA	Assisting Hand Assessment
IG	Intervention group
CG	Control group

SUMMARY

INTRODUCTION

The term cerebral palsy (CP) describes a group of deficiencies in the development of movements and postures attributed to non-progressive disorders that occurred in the immature brain (Rosenbaum et al., 2007). Although the brain damage is not progressive, the clinical manifestations of CP may develop over time and secondary complications may arise as the child ages (Menkes & Sarnat, 2000). Motor disorders in CP are often associated with disorders of sensation, perception, cognition, communication and behavior, epilepsy, and secondary musculoskeletal problems (Krigger, 2006). CP is one of the most common causes of motor disorders in childhood, affecting about 2 to 3 per 1,000 children born alive (Aravamuthan & Waugh, 2016; Bax, Tydeman, & Flodmark, 2006). In underdeveloped countries, its prevalence may reach 7 per 1,000 live births (Taub, Ramey, DeLuca, & Echols, 2004).

Among the different types of CP, Hemiplegic Cerebral Palsy (HCP) is one of the most frequent manifestations, present in 30 to 40% of cases (Sellier et al., 2016). CP is characterized by unilateral involvement of the upper and lower extremities, contrary to the injured cerebral hemisphere (Mewasingh et al., 2004). According to neuroimaging studies, the most frequent causes of HCP are perinatal stroke and congenital malformations (Nelson, 2008). Clinical manifestations of HCP include the classical signs of upper motor neuron injury (pyramidal system), such as muscle weakness, changes in muscle length and recruitment, spastic hypertonia, and hyperreflexia (Jones et al., 2007; Morris, 2007). The delay in the acquisition of motor abilities, as well as deficits in the coordination of body movements of affected extremities, is also observed in children with HCP (Rosenbaum, 2007). In addition, children with HCP tend not to use the affected upper extremity in bimanual tasks of daily life, resulting in learned disuse (Fontes et al., 2016; Steenbergen, Jongbloed-Pereboom, Spruijt & Gordon, 2013). This failure to spontaneously use the affected upper extremity is known as "developmental disregard" and is a major cause of functional limitation (Houwink et al., 2011).

Children with HCP have several motor deficits, however, besides the classic pyramidal deficiency, its clinical manifestations also suggest impairments in the extrapyramidal and cerebellar systems. There is evidence that children with HCP have deficits in motor imagery (Mutsaarts, Steenbergen, & Bekkering, 2007); deficits in body representation and perception (Fontes et al., 2016); presence of involuntary movements (Klingels, et al, 2016); slowness in movements (Steenbergen et al, 2000); dystonia (Gordon, & Duff, 1999); motor incoordination (Eliasson, & Gordon, 2000); balance deficits (Bonan et al, 2004); abnormal gait patterns (Buckon et al., 2001); visuospatial deficits (Hawe et al., 2020); proprioceptive deficits (Kuczynski, Dukelow, Semrau, & Kirton, 2016). Thus, it is important that the child with HCP undergoes a complete neuromotor exam to identify neuromotor impairments at different levels of motor system integration (pyramidal, extrapyramidal and cerebellar).

A standardized neurological examination provides additional support for functional diagnostic performance, allows the identification of pathophysiological mechanisms and helps to define the prognosis, monitor longitudinal history and even evaluate the effects of interventions (Feys & Lisa, 2020). According to Tavano et al (2010), the results of the neuromotor exam allow the identification of dysfunctions in one or more of the three levels of motor integration in the brain: ii) extrapyramidal or basal movements (involuntary movements); and iii) cerebellar (motor coordination, balance, etc.). Despite this evidence, most of the studies that evaluated the neuromotor symptoms in HCP focused on identifying symptoms related to the pyramidal system, such as strength, hyperreflexia, flexibility, tonus, among others. There are few studies involving children with HCP that evaluate non-pyramidal neuromotor signs. Moreover, efforts to interpret neurological signs in HCP in terms of its association with different levels of motor system integration have not been made.

Another aspect little investigated in the literature is the detection of sensorimotor deficits in the non-paretic limb of children with HCP. Motor impairments have been well documented in the upper hemipartic extremity, however, it is unclear whether early unilateral injury resulting from HCP can impair typical non-paretic hand function (Steenbergen & Meulenbroek, 2006). Although the non-paretic hand function in HCP has been considered normal for many years (Gordon & Duff, 1999), more recent studies have shown divergent results. Studies involving the non-plegic hand of children with HCP, reported deficits in coordination, dexterity, strength, and speed of movement (Hawe et al., 2020; Rich et al., 2017; Tomhave, Van Heest, Bagley, & James, 2015). These deficits in the non-paretic hand are often masked by the complex clinical presentation of the affected hand and negatively influence the child's ability to engage in bimanual tasks (Rich et al., 2017).

A possible explanation for the sensorimotor impairments observed in both hands of children with HCP are deficits in motor planning. A large number of studies have shown that the compromised bimanual performance experienced by children with HCP is not only due to difficulties in motor execution, but may also be the result of a deficiency in action planning (Krajenbrink, Crichton Steenbergen, & Hoare, 2019; Lust, Spruijt, Wilson, & Steenbergen, 2018; Steenbergen et al., 2013; Mutsaarts, Steenbergen & Bekkering, 2006). As proposed by Johnson-Frey, McCarty, and Keen (2004), motor planning can be defined as the ability to anticipate the end of a motor action by preparing a certain movement toward an object. Thus, motor planning is associated with the internal action control model proposed by Wolpert (1997). The internal motor control model is a neural system that simulates the next action. This model acts as a predictor in the central nervous system (CNS), providing predictions that allow the successful planning and execution of the action (Wolpert, 1997; Frith & Wolpert, 2000). For each predicted action, the CNS issues a motor command to the muscles and, simultaneously, a copy of the motor command is used to predict the future state of the limb in motion (Frith, Blakemore & Wolpert, 2000; Wolpert & Flanagan, 2001). Hence, motor planning is important because it follows the intentions and plans of the motor acts, assessing whether the actions taken correspond to the desired actions.

For Steenbergen, et al. (2009), despite evidence of motor planning deficits in HCP, current rehabilitation techniques are predominantly focused on relieving motor performance deficits, while motor planning or preparation processes are not addressed. For example, constraint-induced movement therapy focuses predominantly on the affected arm (Chiu, & Ada, 2016). Therapies in which both upper limbs are trained, are also applied to children with HCP, such as bimanual training - HABIT (Friel, et al., 2016). Although sequential action training is included in both of the treatment protocols mentioned above, motor planning is not explicitly trained (Steenbergen, et al., 2009). Evidence suggests that motor planning problems can also significantly affect the performance of daily life activities and therefore need to be addressed (Steenbergen et al., 2013).

The research suggested motor imagery (MI) as a promising method to reduce motor planning deficits observed in children with HCP (Adams, Lust, & Steenbergen, 2018; Williams, Reid, Reddihough & Anderson, 2011; Steenbergen et al., 2009). MI refers to the ability to imagine a motor action without the explicit execution of movement (Decety & Grezes, 1999). More precisely, MI is a cognitive process in which a given motor act is performed internally, through working memory, without an external output to the motor act (Jackson, Lafleur, Malouin, Richards, & Doyon, 2001; Jeannerod, 2001). According to Caeyenberghs, Tsartos, Wilson and Smits-Engelsman (2009), MI plays an important role in effective motor action planning, providing a window to the neurological processes involved in representing actions. MI is able to recruit the aspects involved in the internal action control model to predict the sensory consequences of imagined actions (Kilteni, Andersson, Houborg, & Ehrsson, 2018). Furthermore, MI shares neural substrates similar to those involved in motor planning and execution (Sharma & Baron, 2013). Brain regions, such as the supplementary motor area (Grezes, & Decety, 2001), pre-motor cortex (Jackson et al., 2001), primary motor cortex (Gerardin et al., 2000), cerebellum (Lotze et al., 1999) and posterior parietal cortex (Grezes, & Decety, 2001), are activated during the execution and imagination of different motor actions. Thus, when executing MI, the individual engages neural networks involved in the motor act, keeping the neural system active, which will make it easier to perform the physical movement.

MI training has proven to be effective for learning and optimizing general motor performance and sports skills (Gentili, Papaxanthis, & Pozzo, 2006). In recent years, the application of MI as a tool for motor function recovery has been effective in the recovery of upper limbs, post-accident (Kho, Liu & Chung, 2014), in children with developmental coordination disorder - DCD (Wilson et al. , 2016) and in children with diplegic CP (Cabral, Narumia, & Teixeira, 2010). In children with HCP, Cabral-Sequeira, Coelho & Teixeira (2016) showed positive results with the use of MI to learn a specific motor task. Thus, it would be reasonable to suggest that MI training can be an effective complement to upper limb rehabilitation in HCP. In these children, not only motor execution with the plegic hand is impaired, but motor planning is also compromised, affecting action performance with both hands (Steenbergen et al., 2009). These motor planning deficits can be addressed with MI training. Despite this evidence, the use of MI in the rehabilitation of upper limbs of children with HCP has not been explored extensively.

Thus, based on the identification of this gap in the literature, a study capable of examining motor function in children with HCP with a comprehensive neuromotor exam was performed, considering the three levels of integration specified above. We also investigated whether the early brain damage resulting from HCP affected the non-paretic upper extremity. We then verified the development of MI in children with typical development, determining the age at which MI can be performed. Subsequently, we clarify whether children with HCP are capable of performing MI tasks, and finally, we provide preliminary data on the effectiveness of MI training in the rehabilitation of upper limbs of children with HCP.

In this study, we developed a training protocol based on MI, aiming at the motor recovery of upper limbs of children with HCP. Therefore, before conducting the experiment involving MI in children with HCP, some gaps should be clarified. First, it is not clear if the neuromotor impairments present in children with HCP occur at all levels of motor system integration (pyramidal, extrapyramidal, and cerebellar). Second, we need to check whether the neuromotor changes resulting from HCP also affect the non-paretic limb. Third, we need to establish at what age children can already be involved in MI tasks. To effectively apply MI training in HCP, it is pertinent, first, to establish the age at which children are actually capable of performing this ability. Despite the fact that some studies have been developed, the results are convergent and do not allow a definitive conclusion (Spruijt, Van der Kamp & Steenbergen, 2015; Hoyek et al., 2009; Molina, Tijus & Jouen, 2008; Funk, Brugeer & Wilkening, 2005). Finally, it is necessary to establish whether, even after a premature injury, children with HCP can perform tasks involving MI. Motor planning is compromised in HCP (Steenbergen et al., 2013). Because MI and motor planning share similar neural substrates, it is possible that the motor planning deficiency observed in this population may be related to a reduced capacity to use MI (Mutsaarts, Steenbergen & Bekkering, 2006).

Dissertation Structure

Following the recommendations of the Postgraduate Program in Neuroscience of the Universidade Federal de Minas Gerais - UFMG, this work will be presented in the form of scientific articles. Thus, the thesis is composed of four empirical articles.

In Study 1, after checking the validity and reliability of a comprehensive neuromotor exam, motor function was examined in children with HCP in order to identify whether neuromotor deficiencies were associated with the three levels of motor system integration, i.e., pyramidal, extrapyramidal and cerebellar. It was also investigated whether neuromotor disorders are present in both upper extremity of children with HCP. Study 1 was entitled “Bilateral impairments at different levels of motor integration in hemiplegic cerebral palsy” and will be submitted to the journal *Pediatric Neurology* .

In Study 2, differences in MI development in school-age and preschool children (ages 6-13) were investigated to determine at what age they are able to engage in tasks that require the use of MI. It was also verified whether the ability of MI changes with increasing age. This study was entitled “Motor imagery development in children: Changes in speed and accuracy with increasing age” and was published in the journal *Frontiers in Pediatrics* (DOI: 10.3389/fped.2020.00100).

In Study 3, it was investigated whether children with HCP are able to perform an MI task by comparing their performance with that of healthy controls. Additionally, in this study we investigated whether the ability of MI is associated with the functional and cognitive ability of children with HCP. Study 3 was entitled “Ability of children with cerebral palsy to use motor imagery follows mechanical constraints and does require working memory” and is under review in the journal *Developmental Medicine & Child Neurology*.

In Study 4, a quasi-experimental project was developed to preliminarily investigate the feasibility of using MI training to improve upper limb functional performance in children with HCP. This article was published in the journal *NeuroRehabilitation* (DOI: 10.3233/NRE-192931) and was entitled “Effect of motor imagery combined with physical practice on upper limb rehabilitation in children with hemiplegic cerebral palsy”.

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AIMS

General objectives

(a) To examine neuromotor function in children with HCP using a comprehensive neuromotor exam capable of identifying impairments in both upper limbs, considering the three levels of integration of the motor system (pyramidal, extrapyramidal and cerebellar).

(b) To investigate the development of MI ability in children with typical development, identifying possible changes as age increases.

(c) To evaluate MI ability in children with HCP and its association with motor and cognitive abilities.

(d) To evaluate the adequacy and make a preliminary analysis of the effectiveness of interventions based on MI in the rehabilitation of the functions of the upper limbs in children with HCP.

Specific objectives

(a) To evaluate the validity and reliability of a neuromotor exam for children with HCP.

(b) To verify whether neuromotor impairments in children with HCP will be observed at the three levels of brain integration (pyramidal, extrapyramidal, cerebellar).

(c) To verify whether children with HCP have neuromotor impairments in the non-plegic limb.

(d) To evaluate whether the results of the neuromotor exam of children with HCP can be influenced by the laterality of the lesion.

(e) To verify whether children aged 6 to 7 years old are able to engage in a task that requires the use of MI ability.

(f) To investigate whether the MI ability in children with typical development improves with increasing age.

(g) To identify at what age the ability of MI in children with typical development is equivalent to the ability of healthy adults.

(h) To compare MI ability in children with HCP compared to children with typical development.

(i) To verify if the laterality of the brain injury can influence the MI ability in children with HCP.

(j) To identify possible association between MI ability and the variables functional performance, working memory, intelligence and age in children with HCP.

(k) To verify if an intervention protocol using MI is feasible to improve the functional performance of the upper limbs of children with HCP.

STUDY 1: BILATERAL IMPAIRMENTS AT DIFFERENT LEVELS OF MOTOR INTEGRATION IN HEMIPLEGIC CEREBRAL PALSY

Unpublished manuscript

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Abstract

Introduction: Although Hemiplegic Cerebral Palsy (HCP) results in damage to the unilateral pyramidal system, the clinical presentation of neuromotor deficits is also suggestive of extrapyramidal and cerebellar symptoms bilaterally. This study aimed to establish a neuromotor exam protocol capable of assessing impairments at the three levels of motor integration of children with HCP. The presence of impairments in the non-paretic upper limb was also evaluated, as well as the influence of the laterality of the lesion on the results of the neuromotor exam. **Methods:** The study evaluated 30 children with HCP (10.79 ± 2.61 years) and 60 healthy children (8.27 ± 1.57 years). The children were submitted to intelligence assessment and classified by the manual ability classification system (MACS). A neuromotor exam protocol was developed with specific tasks for each level of motor integration: pyramidal, extrapyramidal and cerebellar. **Results:** The groups did not differ with respect to intelligence ($p = 0.256$) and sex (p

= 0.758) but differed for age (p <0.001). The internal consistency was good for the tasks of the pyramidal and cerebellar levels (> 0.84), but it was low for the extrapyramidal level (0.41). Most neuromotor tasks correlated with MACS (p <0.05). Controls were significantly superior to children with HCP for neuromotor tasks at different levels of integration (p <0.05). The performance of the non-plegine hand in the HCP was significantly inferior to the controls (p <0.05). With the exception of a fine motor ability task, children with right and left HCP did not differ. **Conclusion:** Children with HCP show neuromotor signs suggestive of impairments in the three levels of integration of the motor system bilaterally, although more subtle in the non-paretic limb. For the sample of children in this study, the laterality of the brain injury did not influence the results of the neuromotor exam.

Keywords: cerebral palsy, hemiplegic, neuromotor exam, Ipsilateral side,

Introduction

Cerebral palsy (CP) is one of the most common causes of motor disability in childhood, affecting about 1.2 to 3.6 per 1.000 children born alive (Lang et al., 2012; Aravamuthan, & Waugh, 2016). It is a condition caused by non-progressive multifactorial damage that affects the developing central nervous system, resulting in a set of disorders in movement and posture (Rosenbaum, 2007). Motor deficits in CP are often accompanied by sensory, perceptual, cognitive deficits and learning disorders, among others (Fluss & Lidzba, 2020, Fontes et al., 2016; Rethlefsen, Ryan, & Kay , 2010; Morris, 2007).

CP can be classified into 3 subtypes according to the clinical presentation of their primary motor deficits (Jones, Morgan, Shelton, & Thorogood, 2007; Menkes & Sarnat, 2000). The spastic subtype is the most common (approximately 70 to 80% of cases) and results from lesions in the pyramidal system, that is, in the corticospinal pathways of the brain (Jones, Morgan, Shelton, & Thorogood, 2007). Spastic CP is mainly manifested by spastic hypertonia, hyperreflexia and muscle weakness (Morris, 2007). In addition to the spastic type, CP can be differentiated into dyskinetic (approximately 10 to 15% of cases) and ataxic (approximately 5% of cases). Dyskinetic CP occurs due to lesions in the base ganglios (extrapyramidal system) and is manifested by dysregulation of muscle tone and involuntary movements (Sanger et al., 2003; Jones et al., 2007). Finally, ataxic CP results from cerebellar lesions and presents symptoms such as motor incoordination, dysmetria, imbalance and postural instability (Musselman et al., 2014;

Jones et al., 2007). CP may also be classified according to the topographic pattern of involvement of the limbs as quadriplegic, diplegic and hemiplegic (Jones et al., 2007).

Hemiplegic Cerebral Palsy (HCP) is one of the most frequent manifestations of CP, present in approximately 30 to 40% of cases (Sellier et al., 2016). HCP is characterized by unilateral spastic paresis or plegia attributable to contralateral brain injury (Mewasingh et al., 2004). Children with HCP generally exhibit a delay in the acquisition of motor landmarks, as well as deficits in the coordination of body movements of the upper and lower extremities contralateral to brain damage (Rosenbaum, 2007). As described by Fontes et al., (2016), children with HCP often disregard or do not use the affected upper limb in bimanual tasks, and are unable to involve the affected limb as support for the non-paretic limb. This failure in spontaneous use of the affected upper extremity is known as "developmental disregard" and is an important cause of functional limitation (Houwink et al., 2011). To document and classify the functionality of these children's upper limbs different scales are used. Currently, the Manual Ability Classification System - MACS (Eliasson et al., 2006) is the most widely used and describes how children use their hands to manipulate objects in daily activities.

Although HCP results from damage to the pyramidal system, the clinical presentation of sensorimotor deficits is also suggestive of extrapyramidal and cerebellar symptoms. Fontes et al., (2016) observed deficits in body representation and perception in children with HCP. Klingels, et al. (2016) reported the presence of involuntary movements. In addition, there is evidence for slowed movements (Steenbergen et al., 2000), balance deficits (Bonan et al., 2004), abnormal gait patterns (Buckon et al., 2001) motor incoordination (Eliasson, & Gordon, 2000), and dystonia (Gordon, & Duff, 1999). Thus, it is important that children with HCP undergo a comprehensive neuromotor exam to identify sensorimotor impairments at different levels of motor system integration (i.e., pyramidal, extrapyramidal and cerebellar).

Different neurological tests capable of assessing neuromotor functioning have been suggested. Currently the most widely used neuromotor exams are Physical and Neurological Exam for Subtle Signs (PANESS) proposed by Denckla (1985), exam of minor neurological dysfunctions (Groningen assessment) proposed by Touwen & Prechtl (1970), and Zurich Neuromotor Assessment (ZNA), proposed by Largo et al. (2003). These tests focus primarily on minor neurological dysfunction. These include involuntary or stereotyped movements, dysmetria, tremor, postural changes, incoordination, etc. (Larson, et al., 2007). In general, the presence and severity of such neurological signs allows conclusions on severity of the inferred brain damage. In addition to inferences about the presence of any damage to the developing nervous system, these indices provide valuable evidence to help identify simultaneously compromised behavioral and cognitive mechanisms (Hadders-Algra, 2002).

Most often, MRI is used to identify abnormalities in the brain and is considered the gold standard to support clinical presentation and guide treatment (Staudt, 2010). However, it is a high-cost test, making it difficult to access for populations with low socioeconomic status. Neuromotor exam may be an approach for specifying the location of the lesion or dysfunction in children with HCP. It is a fast, inexpensive, non-invasive and effective clinical approach to identify possible sequelae of diseases that have affected the developing brain (e.g., Denckla, 1985). The establishment of a standardized neurological exam may provide additional support for functional diagnosis (Andrade et al., 2012a,b), allow to identify physiopathological mechanisms to better understand disorders, help define prognosis, monitor the longitudinal history, and document effects of interventions (Feys & Lisa, 2020).

To date, the literature on CP has paid relatively little attention to the interpretation of clinical-anatomical correlations of observed neuromotor disorders. Studies of children with HCP and other neurodevelopmental disorders often determine only the presence and severity of motor difficulties, focusing on the affected upper extremity (Rich et al., 2017). More exhaustive examinations are rarely performed to interpret the results of the neuromotor exam in terms of localization of brain damage. Effort should be invested in analys in correlations among neurological signs observed in the motor exame and their possible neuroanatomical correlates.

A notable exception to this rule are the studies by Tavano et al. (2010) and Gagliardi et al. (2007). Tavano et al examined a sample of individuals with Williams syndrome and observed two specific types of motor impairment. A subtype was characterized by deficits such as incoordination, dysdiadokokinesia, tremor, changes in postural reflexes and ataxia, which point to cerebellar impairments. In contrast, the other subtype presented with choreic form movements, abnormal involuntary movements, grimaces, dystonia and abnormal postures, as main characteristics, which is suggestive of extrapyramidal involvement. In a four-year follow-up study involving individuals aged 3 to 30 years, Gagliardi et al found an age-related neuromotor pattern in individuals with Williams syndrome. In this study, symptoms associated with extrapyramidal involvement became more evident from the age of 8 years and increased in the age group up to 14 years. Symptoms indicating cerebellar impairments were frequent in the sample from childhood to adulthood.

Another aspect explored only rarely in the literature so far is the detection of more subtle neurological symptom of sensorimotor dysfunction in the HCP, in particular in the non-paretic limb. Although motor deficits of the contralesional upper limb are well established in HCP and are the main focus in rehabilitation, few experimental studies have examined motor capacity of the non-paretic upper extremity (Steenbergen & Meulenbroek, 2006). As such, it is not clear whether the early unilateral brain damage caused by HCP can impair the typical functioning of both upper extremities. Staudt (2010) reviewed the development of neural connections in the contralesional and ipsilesional pyramidal tract in individuals with HCP. It is known that perinatal brain injury can disrupt the typical course of brain functioning (Eyre, 2007). And children with HCP do not always show a typical pattern of contralateral reorganization (Staudt et al., 2004). Cortical projection patterns may also be reorganized and occur in an ipsilateral or mixed way (Staudt, 2010; Eyre, 2007; Staudt et al., 2004). Functioning of the non-paretic hand in HCP has long been considered within the norm for children with typical development (Gordon & Duff, 1999). However some more recent studies reported deficits in coordination, dexterity, strength and speed of movement for the non-paretic hand in children with HCP (Hawe et al., 2020; Rich et al., 2017; Tomhave, et al; 2015). These deficits of the non-paretic hand are often masked by the complex clinical presentation of the affected hand (Rich et al., 2017). Substantiating that the functioning of the upper limb on the non-plegic side is preserved in HCP is of particular importance, because that side is often used in a compensatory manner to perform daily tasks (Steenbergen & Meulenbroek, 2006). In addition, most functional tasks, such as activities of daily living, depend on the contribution and coordination of both arms to be successfully performed (Kuczynski et al., 2018).

Despite not presenting neuroimaging data, Tavano et al. (2010) suggested that the results of their neuromotor exam may be interpreted as a result of dysfunctions at one or more of the three levels of motor integration in the brain: i) pyramidal or cortical (fine motor skill, apraxia); ii) extrapyramidal or basal movements (involuntary movements); and iii) cerebellar (motor coordination, balance, etc.). To date, most studies that evaluated neuromotor signs in HCP focused on identifying symptoms related to the pyramidal system, such as strength, hyperreflexia, flexibility, tone, among others. In addition, assessment largely focused on the affected upper extremity.

In this study, we follow Tavano et al. (2010) to evaluate motor function in children with HCP with a comprehensive neuromotor exam, considering the three levels of motor integration as described above. For this, we evaluated the validity and reliability of the neuromotor exam by means of Cronbach's alpha. We also correlated the results of the neuromotor exam with children’s MACS scores to investigate whether the early brain injury resulting from HCP affected the non-paretic upper extremity. Finally, we assessed whether the laterality of the lesion influenced the outcome of the neuromotor exam of children with HCP. In particular, we evaluated the following hypotheses: a) We expected the neuromotor exam to have good validity and reliability; b) Neuromotor deficiencies were expected to be observed at all three levels of motor integration (i.e., pyramidal, extrapyramidal, and cerebellar) in children with HCP with different combinations; c) Neuromotor impairments in the non-paretic upper limb were expected to be seen in children with HCP; and d) Neuromotor impairments the non-paretic limb should not be affected by the laterality of brain damage.

For this, we developed a neuromotor exam protocol, specifically considering the three levels of motor integration (i.e., pyramidal, extrapyramidal, and cerebellar) and applied it to a group of 30 children with HCP, comparing their performance to that of 60 healthy children from a control group with similar cognitive developmental levels and socioeconomic conditions.

Materials and Methods

Participants

The study included a convenience sample of 30 HCP children with a mean age of 10.79 ± 2.61 years, recruited in nongovernmental organizations that provide remediation services for unprivileged children. Participants with HCP were divided into two groups according to the laterality of the brain injury: right hemiplegic cerebral palsy (RHCP), composed of 16 (mean age = 10.50 ± 2.57 years) and left hemiplegic cerebral palsy (LHCP) with 14 children (mean age = 11.17±2.30 years). Inclusion criteria were: individuals diagnosed with HCP aged between 6 and 14 years, with preserved cognitive functioning, absence of intractable epilepsy and clinically relevant psychopathological disorders. The control group comprised 60 children with no history of neurosensory and neuropsychiatric disorders (mean age = 8.27 ± 1.57 years). All children came from a similar socio-economic and cultural background, as they attended free of charge state-runned schools and non-governmental rehabilitation facilities. Participants with HCP were comparable with controls in mental age, asserted through Raven's CPM test.

Ethics

All research procedures were approved by the Research Ethics Committee of the Federal University of Minas Gerais (CAAE: 39793614.7.0000.5149). Written informed consent was obtained from parents or legal guardians of all children prior to assessment. Furthermore, participation was dependent on children's oral consent.

Behavioral Assessment

Hand lateral dominance was assessed using the Laterality Task (Lefevre, 1972). General cognitive abilities were assessed using Raven's Coloured Progressive Matrices test (Angelini et al., 1999). Additionally, participants with HCP had their manual ability classified with the MACS (Eliasson et al., 2006).

Neuromotor exam

The neuromotor exam was designed to assess motor functions at all three levels of motor integration, as suggested by Gagliardi et al. (2007, see also Tavano et al., 2010). Specific tasks were selected to assess different functions indicative of the pyramidal, extrapyramidal and cerebellar level, respectively (see Table 1 for a overview of tasks including brief descriptions). The neuromotor exam was applied individually in a quiet place in order to avoid distractions by children.

Table 1: Description of the tasks of the neuromotor exam.

Level of integration	Task	Operationalization	Coding	Reference
Pyramidal	Nine-Hole Peg Test	Timed removal and insertion of 9 pegs in a pegboard. Each hand tested twice.	Average execution time for each hand in seconds.	Poole et al., 2005
	Finger Tapping	Timed tapping in a pocket calculator (1+1) for 10 seconds. Each hand tested twice.	Average number of taps for each hand.	Davis & Dean, 2011.
	Imitation of Meaningful Gestures	Imitation of 10 meaningful hand gestures (e.g., sending a kiss, "OK" sign, etc.) presented as animation on a computer screen. Instruction to correctly imitate the gesture, regardless of laterality.	Number of meaningful gestures correctly imitated.	Fontes el al., 2014.
	Imitation of Meaningless Gestures	Imitation of 10 meaningless gestures (e.g., fingers in different configurations [k = 5], different configurations of hand in relation to trunk/head [k = 5]) presented as static images on a computer screen. Instruction to correctly imitate the gesture, regardless of laterality.	Number of meaningless gestures correctly imitated.	
	Coin task	To pick up a coin from a table. Each hand tested twice.	Unimpaired: finger grip to pick up the coin. Impaired: dragging the coin from one hand to the other.	Binkofski & Fink, 2005, Platz, 2005.
Extrapyramidal	Motor Sequence	Imitation of three sequenced hand movements performed by the examiner (fist-palm-edge) for six consecutive trials.	Unimpaired: performance in the correct order. Impaired: perform in incorrect order.	Dubois et al., 2000
	Conflicting Instructions	Child taps once when examiner taps twice and child taps twice when examinar taps once.	Unimpaired: ability to follow the contingency Impaired: inability to follow the contingency	
	Go no Go	Child taps once when the examiner taps once. Child does not tap when the examiner taps twice.		
	Motor persistence	Child stretches arms forward with palms down, keeping tongue out and closing eyes for 20 seconds.	Unimpaired: Maintenance of posture. Impaired: No support of posture, involuntary movements, synkinesis and choreic hand.	Gagliardi et al., 2007.
Cerebellar	Index-index	Child accurately touching the tip of the index finger with the tip of the examiner's index several times	Unimpaired: Accurate execution. Impaired: Dysmetria or tremor.	Hadders-Algra, 2010.
	Dysdiadokokinesia	Coordinated and alternate hand movements of supination and pronation for 20 trials.	Unimpaired: Correct alternation. Impaired: Inability to alternate.	
	Oseretsky	Coordinated and alternate movements of opening and closing hands with the arms stretched forward for 20 trials.	Unimpaired: Correct alternation. Impaired: Inability to alternate.	
	Romberg	Standing with the heels together, first with the eyes open, then with the eyes closed, for 10 seconds.	Unimpaired: Posture maintaining Abnormal: Postural instability or loss of balance.	
	Tandem posture	Standing with the feet in tandem for 10 seconds.	Unimpaired: Posture maintaining Abnormal: Postural instability or loss of balance.	
	Jumping and clapping	Jumping and clapping twice before returning to the floor. 2 times simultaneously.	Unimpaired: ability to perform. Impaired: inability to perform.	
	Jumping jacks	Jumping with wide spreaded legs and hands clapping overhead once, then returning to standing position with feet together and arms on the sides.	Unimpaired: ability to perform. Impaired: inability to perform.	
	Hand-knee coordination	Alternate movements of tapping left knee with right hand and right knee with left hand.	Unimpaired: ability to perform. Impaired: inability to perform.	
	Balance reaction	Unexpected pushback while standing.	Unimpaired: Maintaining balance. Impaired: Difficulty maintaining balance.	
	Walking on tiptoe	Walking on tiptoe.	Unimpaired: Can perform the task. Impaired: Does not perform the task.	
	Walk on heels	Walking on heels.		

Statistical analysis

All statistical analyses were performed using the SPSS, version 20. Alpha was set to 0.05. The internal consistency of the neuromotor exam tasks was evaluated separately for each level of motor integration (i.e., pyramidal, extrapyramidal, and cerebellar) using the cronbachs alpha. Spearman correlations were run to evaluate between MACS scores and mastery of neuromotor exam tasks. For this, a score was generated summing up the number of categorical tasks mastered successfully a child at each level of motor integration. For the pyramidal level, the score varied could range from 0 to 8, for the extrapyramidal level, the score could vary between 0 and 3, and for the cerebellar level, scores could range from 0 to 11.

Categorical variables were coded as impaired or unimpaired. The approach suggested by Kruskal-Wallis was used to examine between-group differences in interval-scaled variables. To identify significant differences, we performed pairwise comparisons. Chi-square tests were used to compare performance of children with HCP and healthy children on the categorical variables. Fisher's exact test was used to compare performance of children with RHCP and LHCP, as these subgroups were smaller.

Results

Sociodemographic characteristics of participants

The control, RHCP and LHCP groups were homogeneous with respect to the distribution of the sexes (p=0.758), but not regarding to age (p<0.001), with control children being significantly younger than children with LHCP (p<0.028). RHCP and LHCP children did not differ significantly with respect to age (p>0.05). Despite the age difference, it is important to note that children with RHCP and LHCP did not differ from controls in terms of general cognitive ability (see Table 2). Manual ability of the children with HCP was classified at Level II for 43.3%, Level III for 33.3%, and Level IV for 23.3% of children according to the MACS. A total of 78.6% children in the control group were right-handed. In the RHCP and LHCP groups, 100% were left-handed and right-handed, respectively.

Table 2: Characterization of the sample.

		Controls (n= 60)	RHCP (n= 16)	LHCP (n=14)		
		Mean Rank	Mean Rank	Mean Rank	*χ2*	*p*
Age*		36.50	57.76	68.18	23.62	<0.001
IQ*		50.74	42.22	46.74	2.72	0.256
Gender**	*Female*	46.4%	56.2%	58.3%	0.555	0.758
	Male	53.6%	43.8%	41.7%		

* Kruskal-Wallis
**Chi Square for cathegorical variable.
Note: Controls: children with typical development; LHCP: children with left hemiplegic cerebral palsy; RHCP: children with right hemiplegic cerebral palsy.

Validity and reliability of the neuromotor exam.

Internal consistency of the neuromotor exam scores revealed a *Kuder Richardson 20* (KR-20) of 0.842 for the pyramidal level tasks, 0.417 for the extrapyramidal level tasks and 0.879 for the cerebellar level tasks.

In order to verify the validity of the neuromotor exam tasks, we correlated the neuromotor exam tasks with the MACS. Results of the correlation analysis are shown in Table 3. Only the *imitation of meaningless gestures* task and the *pyramidal score* for the dominant hand did not show significant correlations with the MACS ($p > 0.05$). All other tasks correlated significantly with MACS scores both for the paretic as well as the non-paretic hand (all $p<0.05$). Closer inspection of the correlation coefficients indicated that neuromotor tasks that involve execution time, such as *9HPT*, showed a positive correlation with MACS, indicating that the longer the execution time on the task, the worse the level of classification of manual ability. In addition, the *pyramidal score* (non-dominant hand), *extrapyramidal core* and *cerebellar score* correlated positively with the MACS, indicating that the worse the performance on the neuromotor exam, the worse the classification of manual ability. Finally, the *finger tapping* and *imitation of meaningful gestures* showed a negative correlation with the MACS indicating that the better the scores in these tasks, the better the level of manual ability.

Table 3. Correlation Coefficients.

	1	2	3	4	5	6	7	8	9	10	11
1. MACS		.37*	.55*	-.42*	-.73*	-.54*	-.12	.31	.47*	.64*	.52*
2. 9-HPT (non-plegic hand)			.34	-.45*	-.27	-.03	-.27	.38*	.35	.3*	.40*
3. 9- HPT (plegic hand)				-.32	-.41*	-.01	-.10	.01	.10	.36	.28
4. Finger Tapping (non-plegic hand)					.57*	.32	-.34	-.34*	-.33*	-.41*	-.30
5. Finger Tapping (plegic hand)						.18	.27	-.22	-.27	-.74*	-.40*
6. Imitation of Meaningful Gestures							.13	-.08	-.20	-.11	-.45*
7. Imitation of Meaningless Gestures								-.14	-.14	-.24	-.09
8. Pyramidal score (non-plegic hand)									.90*	.28	.20
9. Pyramidal score (plegic hand)										.27	.20
10. Extrapyramidal score											.52*
11. Cerebellar score											

Note: MACS: Manual Ability Classification System; 9-HPT: Nine-Hole Peg Test.

Spearman correlation (* $p<0.05$)

Neuromotor impairments at the three levels of motor integration

Pyramidal level

Results of the comparisons between groups for the tasks of fine motor ability and imitation of gestures are displayed in Table 4. For the *9-HPT* task, children of the RHCP group showed poorer performance than controls, for both the dominant ($p<0.001$) as well as the non- dominant hand ($p<0.002$). Children of the LHCP group performed poorer than controls only for the non-dominant hand on this task ($p<0.001$), as shown in Table 4. Group comparisons for *finger tapping* task resulted in significant differences only for the non-dominant hand, with both HCP groups performing significantly inferior to controls (both $p<0.05$). For *imitation of meaningful gestures* task there were no significant group differences. However for the *imitation of meaningless gestures* task, RHCP children performed significantly poorer than controls ($p<0.032$). In both gesture imitation tasks, both groups with HCP did not differ from each other significantly.

Table 4: Performance in the numerical tasks assessing the pyramidal level

Tasks	Controls (n=60)	RHCP (n=16)	LHCP (n=14)				Controls x RHCP	Controls x LHCP	RHCP x LHCP
	Mean Rank	Mean Rank	Mean Rank	*df*	X^2	*p*	*P*	*p*	*p*
Pyramidal level tasks									
9-HPT (dominant hand)	40.13	66.31	37.50	2	15.25	<0.001	<0.001	0.654	0.002
9- HPT (non-dominant hand)	30.91	76.34	70.01	2	53.81	<0.001	<0.001	<0.001	0.515
Finger Tapping (dominant hand)	55.67	34.50	44.93	2	4.76	0.092	0.098	0.079	1.000
Finger Tapping (non-dominant hand)	54.59	22.31	23.63	2	29.48	<0.000	<0.001	<0.001	0.893
Imitation of Meaningful Gestures	47.58	41.84	32.63	2	4.76	0.090	0.1.04	1.000	1.000
Imitation of Meaningless Gestures	49.13	30.34	40.21	2	7.44	0.024	0.008	0.262	0.350

* p<0.05 (Kruskal-Wallis)

Note: Controls: children with typical development; LHCP: children with left hemiplegic cerebral palsy; RHCP: children with right hemiplegic cerebral palsy.

The results for the analysis of categorical variables for the pyramidal level are shown in Table 5. Fisher's exact test was initially performed for comparisons between groups of children with RHCP and LHCP. For all evaluated tasks, children with RHCP and LHCP did not differ significantly from each other. Therefore, these children were pooled into a joint HCP group which was then compared to the control group. Significant differences were found for all tasks used to assess the pyramidal level, with the control group showing significantly better performance than the HCP group (all $p<0.005$), as shown in Table 5. The results showed that children in the HCP group also differed significantly from controls for the non-paretic hand ($p<0.05$).

Extrapyramidal level

The two groups of children with HCP did not differ significantly from each other on tasks associated with the extrapyramidal level (all $p>0.05$). Therefore, children were pooled into a joint group and then compared to the control group. In the *motor persistence* task, significantly more children with HCP were impaired as compared to the control group ($p< 0.05$, 57,1% vs. 19.6%, respectively). Of the children with HCP, 25% presented *associated involuntary movements*, 14.2% presented *synkinesis*, and 17.9% had *choreic hand*. Results of comparisons between HCP and controls for the motor persistence task are shown in Table 5.

Cerebellar level

Table 5 shows the results regarding the analysis of categorical variables at the cerebellar level. As for the pyramidal and extrapyramidal level, performance on tasks assessing the cerebellar level did not differ significantly between groups of children with RHCP and LHCP ($p>0.005$). Therefore, we pooled these children into a joint HCP group and compared their performance that of the control group. Significant differences were found for all tasks used to assess the cerebellar level, with the control group performing significantly better than the HCP group (all $p<0.005$).

Table 5. Performance in the categorical neuromotor tasks the three levels of integration of the motor system.

Tasks	Controls (n=60)		HCP (n=30)			
	Unimpaired	Impairment	Unimpaired	Impairment	X^2	*p*
Pyramidal level tasks						
Coin task (dominant hand)	100%	0%	82.10%	17.90%	11.360	0.003
Coin task (non-dominant hand)	100%	0%	46.40%	53.60%	38.748	<0.001
Motor Sequency (dominant hand)	91.70%	08.30%	21.40%	78.60%	44.284	<0.001
Motor Sequency (non-dominant hand)	86.70%	13.30%	17.90%	82.10%	39.613	<0.001
Conflicting Instructions (dominant hand)	85.00%	15.00%	57.10%	42.90%	8.154	0.006
Conflicting Instructions (non-dominant hand)	81.70%	18.30%	46.40%	53.60%	5.947	0.016
Go no Go (dominant hand)	96.70%	03.30%	71.40%	28.60%	12.073	0.001
Go no Go (non-dominant hand)	91.70%	08.30%	71.40%	28.60%	6.211	0.017
Extrapyramidal level tasks						
Involuntary movements	93.30%	06.70%	78.60%	21.40%	4.130	0.051
Choreic hand	98.30%	01.70%	85.70%	14.30%	5.673	0.034
Synkinesis	98.30%	01.70%	85.70%	14.30%	5.673	0.034
Cerebellar level tasks						
Index-index	83.30%	16.70%	39.30%	60.70%	17.416	<0.001
Dysdiadokokinesia	90.00%	10.00%	50.00%	50.00%	17.393	<0.001
Oseretski	81.70%	18.30%	57.10%	42.90%	5.947	0.016
Romberg	98.30%	01.70%	75.00%	25.00%	12.577	0.001
Tanden	91.70%	08.30%	10.70%	89.30%	55.680	<0.001
Jumping and clapping	95.00%	05.00%	57.10%	42.90%	19.350	<0.001
Jumping jacks	88.30%	11.70%	67.90%	32.10%	5.381	0.024
Hand-knee coordination	96.70%	03.30%	64.30%	35.70%	16.997	<0.001
Balance reaction	98.30%	01.70%	67.90%	32.10%	17.604	<0.001
Walking on tiptoe	90.00%	10.00%	39.30%	60.70%	25.434	<0.001
Walk on the heels	91.70%	08.30%	46.40%	53.60%	22.24	<0.001

Note: Controls: children with typical development; HCP = Hemiplegic cerebral palsy.

* $p<0.05$.

Discussion

In this study, we developed a neuromotor exam protocol in order to evaluate the presence of deficiencies in the three levels of motor integration (pyramidal, extrapyramidal and cerebellar) of children with HCP. Moreover, our neuromotor exam also considered possible deficiencies of the non-paretic hand in HCP and we also evaluated whether the laterality of brain damage could influence the results. Confirming our initial hypothesis, the results indicate that children with

HCP have deficiencies that suggest impairments in the three levels of motor integration, i.e., pyramidal, extrapyramidal, and cerebellar. We also observed that, for most of the tasks of the neuromotor exam, children with HCP performed worse when they used the non-parasitic hand compared to the controls. Finally, although children with RHCP performed worse in the motor dexterity task (9HPT) with one non-paretic hand (dominant hand), in the other tasks of the neuromotor exam, the RHCP and LHCP groups did not perform differently. In the following section, these results will be discussed and interpreted considering the current literature, as well as their implications for clinical practice and neuro-rehabilitation.

Validity of the neuromotor exam

We assume that the neuromotor test proposed here was valid and reliable. Our results partially support this hypothesis, showing KR-20 values greater than 0.84, suggesting a good internal consistency. However, it is worth noting that the value for internal consistency of tasks at the extrapyramidal level was low (0.41). We believe that this happened due to the small number of tasks used to evaluate this level. We suggest that this flaw can be corrected by adding other tasks to evaluate this level. The literature has suggested tasks such as those which evaluate dystonic movements, stereotypes, choreiform movements, stiffness, among others (Aravamuthan, & Waugh, 2016; Dubois, et al., 1995; Hawker, & Lang, 1990; Albin, Young , & Penney, 1989). Although Tavano et al., (2010) and Gagliardi et al., (2007) did not evaluate the internal consistency of the tasks, they used 5 tasks to evaluate extrapyramidal signs: choreiform movements, movements, involuntary movements, facial faces and stiffness. We believe that adding two or three tasks would solve the problem of low internal consistency.

Our results confirmed that the tasks of the neuromotor exam were mostly correlated with the MACS, confirming a good concomitant validity. MACS is a valid and reliable tool that describes, on five levels, how CP children use their hands to manipulate objects in manual activities in daily life (Eliasson et al., 2006). For children with HCP, MACS classifies their ability to perform tasks, regardless of whether they use one or two hands. As expected, our correlation analyses showed that children with better levels of manual ability (level II at MACS) showed less deficiency in the tasks of the neuromotor exam. Similarly, children with worse grades at MACS (level IV) also showed greater deficiencies in neuromotor tasks. An important point to highlight is that the MACS correlated with the tasks of the neuromotor exam, not only with the paretic hand, but also with the non-paretic hand. This finding confirms the validity of the neuromotor exam and presents a new perspective for a complete evaluation of neuromotor function in children and adolescents with HCP.

Impairment at different levels of motor integration in HCP.

One of the main objectives of this study was to verify if the neuromotor deficiencies in HCP presented at different levels of motor integration. Our results suggest that they did, confirming our initial hypothesis. Although HCP is known for lesions in the pyramidal system (Jones, Morgan, Shelton, & Thorogood, 2007), i.e., lesions in the cortical pathways, our results also show signs of extrapyramidal (basal nuclei) and cerebellar deficiencies. This hypothesis was established based on clinical observations of children with HCP, who also frequently present non-pyramidal symptoms. In general, deficits in motor coordination, balance, dysmetria, involuntary movements, among others, are frequently observed in this population. These results are expected, since there is evidence that the cerebral cortex, especially the frontal cortex, is interconnected with subcortical structures such as the basal ganglia and cerebellum (Hoshi et al., 2005; Middleton, & Strick, 2001; Middleton , & Strick, 2000; Schmahmann, & Pandya, 1997; Alexander, & Crutcher, 1990). Both the basal ganglia and the cerebellum are considered motor structures and influence multiple aspects of motor behavior through their recurrent connections with the motor cortex (Middleton, & Strick, 2001; Alexander, & Crutcher, 1990). According to Hoshi et al. (2005), both structures are interconnected with the cerebral cortex, considering that a large number of cortical neurons project into the basal ganglia, mainly into the flow nuclei and putamen. The projections from the cortex to the cerebellum occur mainly for the pinpoint nucleus (Hoshi et al., 2005). In addition, the projections leave the basal ganglia and the cerebellum and project into regions of the thalamus which, in turn, project again into the cerebral cortex (Percheron et al., 1996). Middleton, & Strick (2001) examined the topographic organization of the entrance of the cerebellum in different regions of the prefrontal cortex and found important findings. First, besides the classical connection to the primary motor cortex, the cerebellum can influence several areas of the prefrontal cortex through its connections with the thalamus. Second, there are output channels in the cerebellum (toothed nucleus) that influence both motor and cognitive functions. Finally, the authors suggested that the cerebellar projections in the prefrontal cortex may also be related to motor planning and work memory functions. Middleton, & Strick (2000) also confirmed projections of the basal ganglia and cerebellar outlets to areas of the cortex involved in arm representations: the ventral premotor area and the supplementary motor area. Based on these evidences, a possible interpretation of the origin of the neuromotor deficits found in children with HCP in our study is that they result from an interruption of the connections between the cerebral cortex and the nuclei of the base and cerebellum.

Our results showed that children with HCP have deficiencies in all three levels of motor system integration and raise important questions. So far, assessment tools for children with HCP have focused largely on symptoms indicating pyramidal involvement, such as muscle weakness, spasticity, changes in tone, among others. However, according to our results, there is evidence that the structural abnormalities observed in the brains of children with CP reflect more than focal damage in a single system (Reid et al., 2015; Friel, et al., 2012; Eyre, 2007). It is possible that this focal brain damage may influence the development of other systems, causing additional effects that are more widespread throughout life (Reid et al., 2015; Friel, et al., 2012; Eyre, 2007). Although neuroimaging helps to identify abnormalities in the brain, correlations between the extent of the lesion and functional deficiencies after hemiplegia are low, and individuals with similar brain injuries may have different levels of motor performance (Jette, 1984). Thus, the appropriate rehabilitation program for children with HCP should take into account the clinical symptoms identified during the evaluation and not just the type and extent of any brain damage. It is also important to consider that neuroimaging tests are not accessible to the entire population due to its high cost. The accurate assessment and identification of the degree of motor disability enables the development of a more effective treatment strategy and is therefore a key factor in the rehabilitation of these children. As a result, a neuromotor exam that assesses neurological symptoms at all levels of motor integration, including symptoms associated with extrapyramidal or cerebellar involvement, is extremely important for the proper selection of the rehabilitation program. The neuromotor test proposed in this study is easy to apply at a relatively low cost and has proven effective in identifying the presumptive level of motor integration at which specific deficiencies occur, thus providing a more realistic understanding of its deficiencies and consequently allowing the selection of more effective interventions.

Children with HCP are a heterogeneous group, with different neuromotor characteristics, which makes it necessary to evaluate them and establish rehabilitation goals individually. Heterogeneity is present both at the level of brain damage observed and at motor levels, with variable severity of sensorimotor deficiencies (Weinstein et al., 2014). Atypical brain development, caused by early brain damage, has a profound impact on development and acquisition of motor skills (Weinstein et al., 2014). Despite this variation in clinical status, most evaluation instruments focus on the function of the affected upper limb. Among the instruments used, the Gross Motor Function Measure - GMFM (Russell et al., 1989) was widely adopted to measure the gross motor function of children with CP. Other instruments, such as the assessment of unilateral upper limb function in Melbourne (Johnson et al., 1994) and the Fugl-Meyer Assessment (FMA scale; Fugl-Meyer et al., 1975) are frequently used in clinical settings to assess upper limb function in patients with HCP (Krebs et al., 2009). However, these instruments are usually based on subjective observational analysis of the patient's ability to perform numerous tasks. On the other hand, the neuromotor exam proposed here is an objective method and seems to be a useful tool to document neuromotor deficits at different levels of motor integration.

Bilateral impairments at HCP

One of the objectives of our study was to investigate the presence of disabilities in the non-paretic hand of children with HCP. To date, few studies have evaluated the motor capacity of the nonparasitic extreme in this population (Steenbergen et al., 2006). In this context, this study adds important information contributing to the hypothesis that a unilateral injury may lead to bilateral motor deficiencies (Kuczynski, et al., 2018; Holmström et al., 2010; Eliasson et al., 2005). In the great majority of the tasks used in our study, including all levels of motor integration, children with HCP presented deficiencies for both hands. These observations are consistent with the results of previous studies (Kuczynski, et al., 2018; Steenbergen et al., 2006). Kuczynski, et al., (2018), for example, reported significant deficits for the non-paretic hand of children with HCP in an outreach task when compared to healthy controls. Steenbergen et al., (2006), in assessing the non-paretic limb in a sample of 5 children with HCP, found worse results in motor dexterity tests and changes in basic kinematic measurements, such as length of motion and hand speed (Steenbergen et al., 2006). Therefore, our results added to these previous conclusions reinforce the hypothesis of bilateral involvement in children with HCP. This is also supported by previous investigations in adults with unilateral post-stroke injuries (Wetter, Poole & Haaland, 2005; Kim, et al., 2003). Wetter, Poole, and Haaland (2005) applied the Jebsen-Taylor Hand Function Test (J-HFT), a widely used tool to simulate daily life activities in post-stroke individuals and found disabilities in the non-plegic hand. In short, these findings contribute to a better understanding of motor deficits in HCP and have important clinical implications for the development and appropriate application of rehabilitation therapies.

Although the brain damage in HCP affects only one of the cerebral hemispheres, our results point to bilateral deficiencies. We believe that a possible explanation for the motor deficiencies observed in both members of children with HCP are deficits in motor planning. Motor planning is related to the ability to anticipate the demands of a task, causing the action to be performed as planned. Motor planning is thus considered a prerequisite for the successful execution of motor actions (Stöckel, Wunsch, & Hughes, 2017; Rosenbaum, Meulenbroek & Vaughan, 2004). Consistent evidence points to commitments in motor planning at HCP (Krajenbrink, Crichton Steenbergen, & Hoare, 2019; Lust, Spruijt, Wilson, & Steenbergen, 2018; Steenbergenet al., 2013; Mutsaarts, Steenbergen & Bekkering, 2006). Krajenbrink et al., (2019) further suggested that impaired motor planning in HCP results in bilateral motor deficiencies. In line with our hypothesis, Steenbergen, Crajé, Nilson & Gordon (2009) showed that deficits in motor planning in HCP can impair performance in daily life, not only in the affected hand, but also when using the non-paretic hand. The researchers found that motor planning deficits observed in HCP can be treated with motor imagery - MI (Mutsaarts, Steenbergen, & Bekkering, 2007). MI, i.e. the imagined movements, share neural networks similar to those involved in motor planning and execution (Sharma & Baron, 2013). Thus, when executing MI, the individual recruits neural networks involved in planning and executing motor acts, which can be an alternative strategy in the motor rehabilitation of children with HCP. Thus, we can suggest that the motor deficiencies observed in both hands of children with HCP are related to deficits in motor planning. These deficits have functional implications, especially in patients who need to use the non-paretic limb to compensate for hemiplegia. Future studies evaluating the efficacy of MI in the treatment of motor planning deficits and, consequently, in the improvement of bimanual function in HCP, are needed.

In our study, the motor deficits in the non-paretic hand were less severe than those observed in the paretic hand. Although to a lesser extent, these deficits can impair performance in almost all tasks of daily life, such as self-care, school, recreation or leisure activities (Sköld et al. 2004). These results have significant clinical implications. Currently, traditional rehabilitation focuses mainly on the treatment of paretic hand deficiencies, reducing the effects of muscle weakness, stiffness and atrophy (Green, & Wilson, 2012). According to Buccino et al. (2012), even when involved in a comprehensive rehabilitation program, including conventional physiotherapy, orthosis use, and spasticity treatment, about 75% of children with HCP may have motor disabilities in daily life activities. By establishing the presence of bilateral motor disabilities in children with HCP, our study contributes to the development and selection of the most appropriate rehabilitation approaches. Currently, the two therapeutic approaches most commonly used in upper limb rehabilitation in children with HCP are: constraint-induced movement therapy (Pierce et al., 2002) and bimanual intensive care - HABIT (Charles & Gordon, 2006). Constraint-induced movement therapy was developed to help patients overcome non-use of the affected arm (Pierce et al., 2002). However, it is potentially invasive, which discourages the use of the non-paretic arm. HABIT, on the other hand, proposes a protocol with activities that require bimanual use (Charles & Gordon, 2006). However, there is currently no consensus on the ideal therapy for individuals with HCP (Rich et al., 2017). Considering that there is a bilateral commitment to HCP in our findings, approaches to encourage bimanual training may be more beneficial to this population.

Does the laterality of the lesion influence motor impairments in HCP?

In this study, children with RHCP and LHCP did not differ significantly in most tasks, with the exception of the motor dexterity task, where children with RHCP performed less well than children with LHCP. These results partially support our initial hypothesis that the laterality of the lesion does not influence the results of the neuromotor exam. In healthy individuals, it is known that the left hemisphere is dominant in motor control (Haaland et al., 2004). A classic example of left hemisphere dominance for action is limb apraxia (Haaland, 2006). Apraxia is commonly seen after left hemisphere stroke in adults and consists of a deficit of skillful movement that cannot be explained by weakness, change in tone or posture, movement disorders, lack of reference, and difficulty in understanding (Koski, Iacoboni, & Mazziotta, 2002; Leiguarda, & Marsden, 2000). Despite the fact that motor functions are predominantly controlled by the left hemisphere, it is possible that perinatal brain damage may interrupt the typical course of brain development (Kuo et al., 2017). Previous studies have shown that different motor projection patterns may develop after early unilateral injury (Staudt, 2010). Some studies suggest that the pattern of motor representation after an early injury is one of the main factors influencing motor development (Klingels et al., 2016; Schertz et al. 2016). Children with HCP may have a contralateral, ipsilateral or even mixed pattern of projection (Kuo et al., 2017; Holmström et al., 2010; Guzzetta et al., 2007). Guzzetta et al., (2007) also suggest that the development of ipsilateral cortical projections is associated with engine malfunction. So, it may be that the pattern of cortical reorganization after an early injury has more influence on the results of the neuromotor exam than the laterality of brain injury. This would explain our findings that the laterality of the lesion did not influence the results of the neuromotor exam of children with

HCP. We think these results are due to the different possibilities of cortical reorganization after injury.

Conclusion

In this study, we were interested in establishing a neuromotor exam protocol that would be able to assess deficiencies in the three levels of motor integration (pyramidal, extrapyramidal and cerebellar) of children with HCP. We found that, in fact, children with HCP present neuromotor signs suggestive of deficiencies in the three levels of motor system integration. The neuromotor exam applied in this study showed that, although more subtle in the non-parasitic limb, motor deficiencies in HCP are present bilaterally. It was also possible to confirm that, for the sample of children in this study, the laterality of brain damage did not influence the results of the neuromotor exam. However, it is worth noting that for fine motor abilities, children with injuries in the left hemisphere showed worse results when compared to children with injuries in the right hemisphere. It is important to mention that due to the limits of the sample size and the extent of our neuropsychological investigation, we will suggest caution in generalizing our results.

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STUDY 2: MOTOR IMAGERY DEVELOPMENT IN CHILDREN: CHANGES IN SPEED AND ACCURACY WITH INCREASING AGE.

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Abstract
Although motor imagery has been pointed as a promising strategy for the rehabilitation of children with neurological disorders, information on their development throughout childhood and adolescence is still scarce. For instance, it is still unclear at what age they reach a development comparable to the motor imagery performance observed in adults. Herein we used a mental rotation task to assess motor imagery in 164 typically developing children and adolescents, which were divided into four age groups (6-7 years, 8-9 years, 10-11 years, 12-13 years) and 30 adults. The effects of biomechanical constraints, accuracy and reaction time of the mental rotation task were considered. ANOVA showed that all groups had the effect of biomechanical restrictions of the mental rotation task. We found a group effect for accuracy (F [4,180] = 17,560; p <0.00; $\eta^2 = 3.79$) and reaction time (F [4,180] = 17.5; p <0.001, $\eta^2 = 0.615$), with the results of children groups 6-7 years and 8-9 years being significantly lower than the other groups (p <0.05). In all the analyses, there were no differences regarding accuracy and reaction time among the participants of the age groups 10-11 and 12-13 years and adults (p> 0.05). Concluding, children aged 6-7 years were able to perform motor imagery, motor imagery

ability improved as the participants' ages increased, and children aged 10 and over-performed similarly to adults.

Keywords: Motor imagery, development, children, adults, mental rotation.

Introduction

The ability to mentally simulate actions without physically performing them is one of the most remarkable skills of the human mind. Motor Imagery (MI) can be defined as a dynamic cognitive process in which an individual mentally simulates an action without the external manifestation of the motor act (Conson, Elisabetta, & Luigi 2013; Jackson et al., 2001). According to Jeannerod (1994) MI is the representation of the action involved in the planning and execution of the movements. Mental simulation of movement is important because it follows the intentions and plans of motor acts, assessing whether the actions performed correspond to the desired actions (Jeannerod, 1994; Wolpert, 1997). Thus, MI exhibits many of the properties of motor planning and is considered a valid method for training the internal action control model (Jeannerod, 1999). The internal motor control model proposed by Wolpert (1997) is a neural system that simulates the next action. This model acts as a predictor in the central nervous system, providing predictions that allow the planning and successful execution of the action (Wolpert, 1997; Frith, Blakemore, & Wolpert, 2000). Thus, for each intended action, the nervous system issues a motor command to the muscles, while a copy of the motor command is used to predict the future state of the moving limb (Frith, Blakemore, & Wolpert, 2000; Wolpert, 2001).

According to Jeannerod (2001), imagined movements are functionally equivalent to those performed physically in terms of intentions, motor planning, and motor program engagement. In fact, functional neuroimaging studies have shown that MI activates a set of neural networks (parietal, frontal motor and cerebellar areas) that partially overlap the brain network that is activated during motor performance (Wriessnegger, Brunner, & Müller-Putz, 2018; Hardwick, Caspers, Eickhoff, & Swinnen, 2017; Kaiser et al. 2014; Grezes & Decety, 2001). Thus, as MI and motor execution are closely related processes, MI is increasingly being explored to improve motor skill acquisition by stimulating the neural networks underlying movement planning and control (MacIntyre et al., 2018; Jackson et al., 2001). Indeed, improvements in the performance of motor skills associated with MI training have been documented in healthy people (Fortes et al., 2019; Weinberg, 2008) and in clinical populations, particularly in post-stroke patients

(Kumar, Chakrapani, & Kedambadi, 2016). Specifically, repetitive activation of neural pathways during MI activates the neuroplasticity mechanisms underlying motor learning, providing a rationale for their use in neuro-rehabilitation. Therapy based on MI and interventions based on the physical practice induce brain plasticity required for functional recovery (Sheahan, Ingram, Žalalytė, & Wolpert, 2018).

To improve motor skills, individuals must imagine all the sensations that accompany the physical performance of the imagined task (Ter Horst, Van Lier, & Steenbergen, 2010). Therefore, determining the extent to which images are used by an individual is critical to ensure the success of the intervention. A variety of MI measurements are available. The vast majority of research involving children uses the mental rotation task or mental chronometry to assess MI ability (Spruijt, van der Kamp, & Steenbergen, 2015; Butson, Hyde, Steenbergen, & Williams, 2012; Caeyenberghs, Tsoupas, Wilson, & Smits-Engelsman, 2009; Kosslyn, DiGirolamo, Thompson, & Alpert, 1998). The present study focuses on the investigation of the capacity of MI using exclusively the task of mental rotation.

Studies that applied the task of mental rotation associated with neuroimaging observe a significant motor activation of the cortex when participants imagined the mental rotation of the hand figures (Kosslyn et al., 1998). In a recent study involving transcranial magnetic stimulation, Hyde et al. (2017) suggest that the motor cortex is activated during the performance of hand laterality judgment task (HLJ). In this task, hand figures are presented in different spatial orientations and individuals mentally simulate the movements of their own hands and decide whether the figures represent the left or right hands. The linear relationship between the angle of rotation and reaction times (RT) proposed by Parsons (Parsons, 1994) was confirmed by studies showing that biomechanical constraints that apply to physical motion also restrict imagined motion (Caeyenberghs et al., 2009). The effect of biomechanical constraints refers to increase in RTs when hand figures are presented in anatomical positions that make mental rotation difficult (Figures with fingers facing sideways). Similarly, a decrease in response time is observed when the stimuli are medially rotated (figures with fingers facing medially). The presence of the effect of biomechanical constraints on the task confirms that individuals indeed used MI (Conson, Elisabetta, & Luigi 2013; Van Nuenen et al., 2012). de Lange et al. (2006) evaluated brain activation of healthy individuals while performing the mental rotation task using functional magnetic ressonance and found stronger activation of pre-motor and intraparietal regions when individuals responded to stimuli presented in medial positions when compared to lateral stimuli.

These findings show that there are indeed differences in judging hand images in medial and lateral postures, therefore providing further support for the hypothesis of the effect of biomechanical constraints.

In addition to changes in RT as a function of the rotation angle, there is a postural effect of the mental rotation task that strengthens the presence of the effects of biomechanical constraints. Thus, the position of the participant's body during the task may influence the recognition of hand laterality (Ter Horst, Van Lier, & Steenbergen, 2010; Lameira et al., 2008; Parsons, 1994). This is because the volunteer simulates the movement of one's body from its current position, and not from a fixed representation in the brain (de Lange, Helmich, & Toni, 2006). To solve the task, the individual keeps his/her hand in the back posture, and therefore shorter RTs for stimuli in this posture are expected than RTs for stimuli presented in the palm view.

Studies involving the adult population established that at this age there is a complete maturation of the mechanisms involved in MI (Iachini et al., 2019). However, there is great controversy as to the minimum age when a child is able to engage in tasks using MI (Iachini et al., 2019; Spruijt, van der Kamp, & Steenbergen, 2015; Conson, Elisabetta, & Luigi, 2013; Butson et al., 2004; Molina, Tijus, & Jouen, 2008). Moreover, the age when they reach development comparable to that observed in adults remains unclear. According to Funk et al. (2005), there are few studies investigating the development of MI. In addition, from studies evaluating MI in children, most compared typically developing children to those with Cerebral Palsy or Development Coordination Disorder - DCD (Barhoun et al., 2019; Lust, Spruijt, Wilson, & Steenbergen; 2018; Jongsma et al., 2016; Lust, Wilson, & Steenbergen; 2016; Deconinck, Spitaels, Fias, & Lenoir, 2009).

From studies that evaluated MI in children using variations of the mental rotation task, some reported the presence of the effect of biomechanical restrictions for children between 5 and 12 years of age (Fuelscher et al., 2016; Spruijt, van der Kamp, & Steenbergen, 2015; Caeyenberghs et al., 2009; Funk, Brugger, & Wilkening, 2005), suggesting that in this age group they are already capable of performing MI based on motor processes. In the study by Funk et al. (2005), about 60% of children aged 5 to 6 years were able to use MI, compared with 100% of adults. However, in a later study, Butson et al. (2014) state that most children aged 5 and 6 years were unable to perform the task accurately above 50% of the correct level. Furthermore, these authors confirmed the presence of the effect of biomechanical restrictions only in children aged 8, 9 and

11 years, in children aged 7 and 10 years, this effect was not found. There is still controversy regarding changes in the effect of biomechanical constraints as age increases. In the study by Funk et al., (2005) the impact of biomechanical constraints and hand posture on solving the mental rotation task was greater in children than in adults, suggesting that children are even more guided by motor processes than the adults. In contrast, this claim was challenged by a later study showing that biomechanical constraints were stronger in 8-year-olds than in 6-year-olds (Toussaint et al., 2013).

Caeyenberghs et al. (2009) compared performance in the mental rotation task of 7- and 8-year-olds, 9- and 10-year-olds, and 11- and 12-year-olds and found that younger children (7 and 8-year-olds) are generally less accurate and slower than older children (11 and 12 years). This finding suggests that there are progressive improvements in MI skills as age increases. In a more recent study, Fuelscher et al., (2016) point to a non-linear relationship between the MI ability and age in the HLJ task. These authors also stated that, in these children from 6 to 12 years old, MI ability is associated with motor planning ability, since they are closely related processes. However, the authors are cautious in interpreting these results in view of the modest sample size.

Taken together, studies of age-related differences in MI indicate that children's ability to accurately perform the mental rotation task increases with age. However, the literature review by Spruijt et al. (2015) suggests that it is not possible to draw definitive conclusions from studies using the mental hand rotation task on the exact development of MI in children. Given the small sample size of the studies, sample error is a major concern and probably contributed to the controversial group comparisons reported in previous studies. Moreover, the limited age ranges proposed by the studies do not allow definitive conclusions about the development of MI in children, its evolution during childhood, adolescence and adulthood.

Given the controversies explicit in the literature, the temporal course of development and the underlying mechanisms have not yet been sufficiently clarified. Involving a larger sample (194 children) and a wider age range (from 6 and 13 years old) than previous studies, and using the mental rotation task herein we investigated: (a) if younger children are already able to perform MI tasks; (b) if children follow the biomechanical constraints to solve the task; (c) if there is influence of postural perspective of the hand: dorsal versus palmar; (d) if there are age-related differences; and (e) at what age children’s MI performance resembles that of healthy adults. To this end, we analyzed the effects of biomechanical constraints on RTs, the effects of back and palm visual perspectives, and the age differences for accuracy and RT.

Materials and Methods

Participants

The total sample consisted of 194 volunteers, of whom 164 are children (88 boys and 76 girls), recruited from a public school in southeastern Brazil (city of Betim, Minas Gerais, Brazil). The ages of the participants ranged from 6 years and 5 months to 13 years and 2 months (mean age = 9.52 ± 2.10 years). Children and adolescents were assembled into four age groups: 6-7 years, 8-9 years, 10-11 years, and 12-13 years old (Table 1). A group of 30 adults was also recruited in Betim, Minas Gerais, Brazil. Only right-handed individuals presenting normal or corrected vision, lack of neuromotor impairment, able to discriminate right and left were included. Before the study initiated, written consent was obtained from the adults as well as from the parents/guardians of the children and adolescents recruited. All research procedures were approved by the Research Ethics Committee of the Federal University of Minas Gerais (COEP / UFMG).

Measurements

Laterality dominance

Lateral dominance of hand was assessed by the Laterality Task (Lefévre, 1976). The participant sat in a chair facing a table. A small ball was placed by the examiner in the center of the table. Then, the participant was instructed to take the ball with one hand and throw it into a basket that was positioned in front of the table. The test was repeated three times. The volunteer who used his right hand to catch and throw the ball in all three attempts was considered right-handed.

Right–left orientation

To evaluate right-left orientation we used the Right–Left orientation test (Dellatolas, Viguier, Deloche, & Agostini, 1998). The test has 12 items of right and left body parts recognition. It is divided into three steps: the first presents simple commands regarding the child’s own body, the second consists of double commands – direct and crossed – toward the child’s body. In the third step, pointing commands to single lateral body parts of an opposite-facing person was issued. Correct answers were scored as one and wrong answers scored as zero.

Motor Imagery

The ability of MI was measured by the hand laterality judgment task (HLJ; Figure 1), which is a variation of the mental rotation task (parsosn, 1994). This is a computerized task in which, on a computer screen, figures of the hands (right and left) are presented in different views (back and palm) and rotation angles (0º, 90º, 180º and 270º). The task consists of 16 different stimuli, repeated five times each, totaling 80 stimuli. The HLJ task evaluates the MI by requiring the individual to imagine his own hand moving to the orientation presented in the stimulus to make the laterality judgment. The use of MI to solve the HLJ task is characterized by differences in RT and accuracy as well as by the presence of the effect of biomechanical constraints (Conson, Elisabetta, & Luigi, 2013; Parsons, 1994). This effect is characterized by an increase in RT as a function of the rotation angle of the stimuli (Kumar, Chakrapani, & Kedambadi, 2016). The stimuli in which the hand figures are medially oriented are anatomically easier to rotate mentally and therefore the resulting RT to recognize medially oriented stimuli should below. Also, judging laterality when the stimulus presented is the left hand rotated 90º (medial rotation) is faster than when the right hand at 90° is shown (de Lange, Helmich, & Toni, 2006; Parsons, 1994).

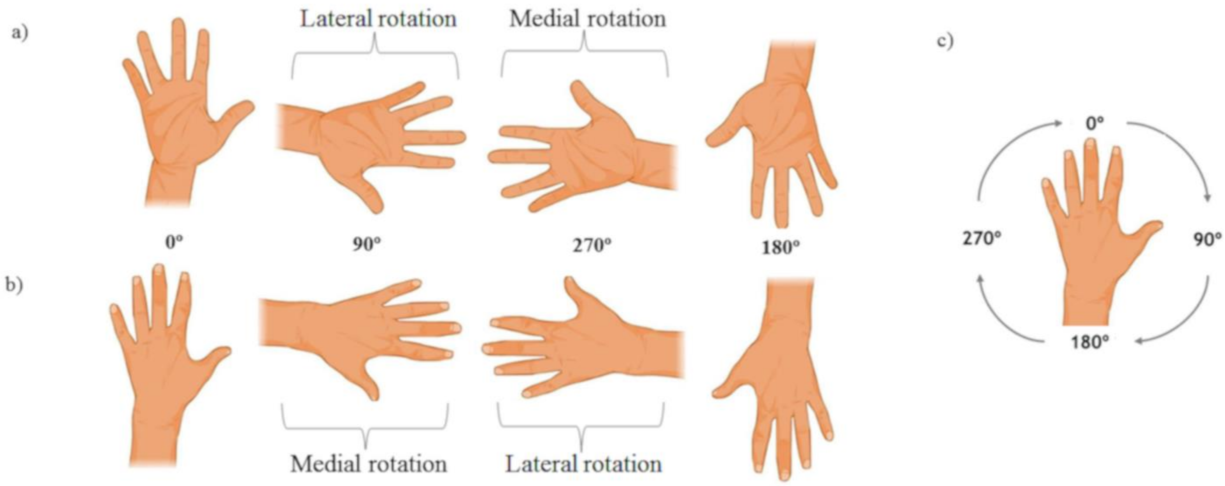

Figure 1: Examples of the hand laterality judgment task stimuli. In (a), the right hand stimuli are observed in palm view. In (b), the left hand stimuli are presented in back view, and in (c) the rotation direction of the task stimuli is indicated.

Procedures

The participants were positioned at a comfortable distance from the computer screen and instructed to decide as quickly and accurately as possible whether each stimulus was a left or a right hand. Hand stimuli were randomly presented at 4 different angles of rotation (using the Presentation software, version 0.71) and remained on the screen until a response was recorded by pressing a designated key on the computer keyboard. Moreover the volunteers were instructed to imagine their own hand turning to the position of the presented stimulus and then decide if the stimulus corresponded to the right or left hand. The literature review by Spruijt et al., (20) states that it is not possible to infer whether or not to use instructions to solve the mental rotation task, due to methodological variations of the studies developed. Thus, based on previous studies (21, 22, 42) our study chose to provide instructions to participants. Participants remained with their hands in the pronated posture (back of the hand up) positioned close to the computer keyboard. Participants were prohibited from moving their hands. The volunteer was instructed to use his/her index fingers to respond by pressing the right computer key with his right finger when the picture was considered to correspond to the right hand and the left computer key when the picture was considered to correspond to the left hand. Accuracy and RT records were produced for each stimulus by and later used for data analysis.

Data analysis

Tests in which participants missed or produced RTs greater than three standard deviations above or below the overall average were excluded from the analyzes. The average time and precision, as well as the average time in medial and lateral rotation for the palmar and dorsal views, were calculated for every participant. To compare the means obtained for accuracy and RTs we performed analysis of variance by the method of the general linear model (ANOVA). For the variables in which ANOVA found significant differences ($p < 0.05$) between the groups, Bonferroni post hoc analysis was used for multiple comparisons. Repeated-measures ANOVA was used to examine the effects of the biomechanical constraints of the HLJ task on RT (angle: medial and lateral; view: dorsal and palmar; hand: right and left). Significant results were analyzed with the t-test for paired samples. Finally, to determine if age predicts efficiency in the MI task, a simple regression analysis was performed.

Results

As shown in Table 1, all five groups had a similar representation of both sexes ($\chi 2 = 0.533$; $p = 0.137$). Nine children were excluded for being left-handed and five were excluded for not being able to discriminate right and left.

Table 1: Sex and age of groups

	Sex		Age	
	Male	Female	M	SD
Group 6-7 years old (n=37)	19	18	6.69	0.48
Group 8-9 years old (n=40)	26	14	8.45	0.53
Group 10-11 years old (n=39)	21	18	10.49	0.65
Group 12-13 years old (n=34)	14	20	12.60	0.51
Group adult (n=30)	13	17	25.77	1.99

M: mean; SD: Standard deviation

Effects of Biomechanical Constraints

Medial rotation versus lateral rotation

Figure 2 shows the presence of the effect of biomechanical constraints, as indicated by ANOVA showing a significant interaction between the rotation angle and RT [$F(4,180)=29.61$; $p<0.006$; $\eta^2=0.580$]. Participants were faster to judge the stimuli presented in medial than in lateral rotations ($p<0.05$). Bonferroni's comparison showed that all age groups were faster to judge medial rotations for both right hand stimuli (6-7 years old: $p=0.001$, $d=2.06$; 8-9 years old: $p<0.000$, $d=1.82$; 10-11 years old: $p<0.001$, $d=1.64$; 12-13 years old: $p<0.013$, $d=1.12$; adult: $p<0.026$, $d=0.98$), and left hand stimuli (6-7 years old: $p<0.001$, $d=2.06$; 8-9 years old: $p<0.000$, $d=1.95$; 10-11 years old: $p<0.001$, $d=1.16$; 12-13 years old: $p<0.013$, $d=0.81$; adult: $p<0.026$, $d=0.86$). We also found a significant interaction between age and RT [$F(4,180)=29.61$; $p<0.006$; $\eta^2=0.580$], with the groups 6-7 years and 8-9 years of significantly slower than the groups 10-11 years, 12-13 years, and adults ($p<0.05$). The other comparisons between the groups did not result in statistically significant differences (Figure 2).

Dorsal view versus palm view

As shown in Figure 2, the ANOVA showed a significant interaction between the rotation angle and the stimulus view [$F(4,180) = 12.81$; $p < 0.001$; $\eta^2 = 0.346$]. Children of the group 6-7 years were only faster to judge dorsal view stimuli for right-hand figures ($p<0.001$, $d=0.68$). The opposite was observed for the left hand, as lower RTs were observed for the palm view ($p<0.001$, $d=-0.22$). Children of the group 8-9 years did not show significant differences to judge back and palm stimuli ([$F(4,180)=2.05$; $p=0.161$; $\eta^2=0.060$]. Pairwise comparisons showed that groups 10-11 years, 12-13 years and adult were faster to judge hand laterality presented in back view, both for the stimuli of the right hand (10-11 years old: $p<0.001$, $d=0.96$; 12-13 years old: $p<0.013$, $d=0.98$; adult: $p<0.026$, $d=1.58$), and left hand (10-11 years old: $p<0.001$, $d=0.80$; 12-13 years old: $p<0.001$, $d=0.54$; adult: $p<0.001$, $d=0.86$).

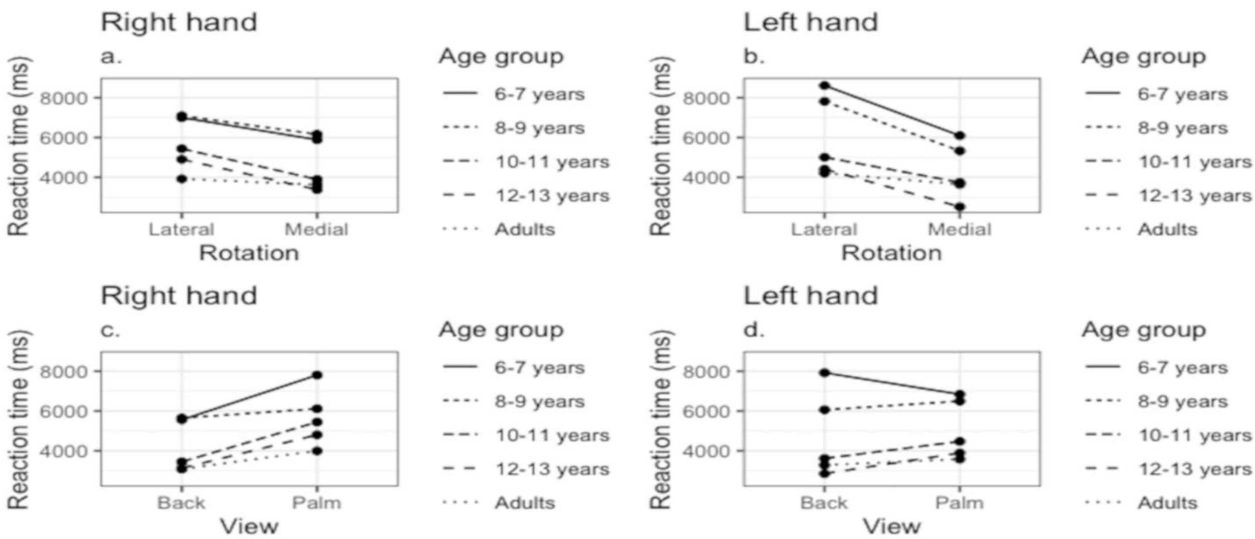

Figure 2: Effect of biomechanical constraints. We verified the reaction time (RT) averages to judge the stimuli in medial rotations compared to lateral rotations for the right hand (a) and left hand (b). We also compared the RT to judge the stimuli presented in the palmar and dorsal views for both the right (c) and left (d) hands.

Age differences

A simple regression analysis revealed that age is a significant correlate of performance in the MI task in terms of accuracy (r^2= 0.357; β = -0.605; t = -6.357; p < 0.001) and RT (r^2= 0.329; β = -0.582; t = -5.982; p < 0.001).

Accuracy

The average of the correct answers (accuracy) is shown in Figure 3. First we confirmed that all participants indeed involved in MI to solve the task by detecting if they responded better than chance (with hit rates above 50%). Accuracy analysis revealed a major group effect (F[4,180]= 17.560; p<0.00; η^2=3.79). The groups 6-7 years and 8-9 years were significantly less accurate than the groups 10-11 years, 12-13 years, and adult group (p <0.05). Groups 6-7 and 8-9 years responded similarly (p> 0.05). In addition, the groups 10-11years, 12-13 years and adult responded similarly in terms of accuracy (p> 0.05).

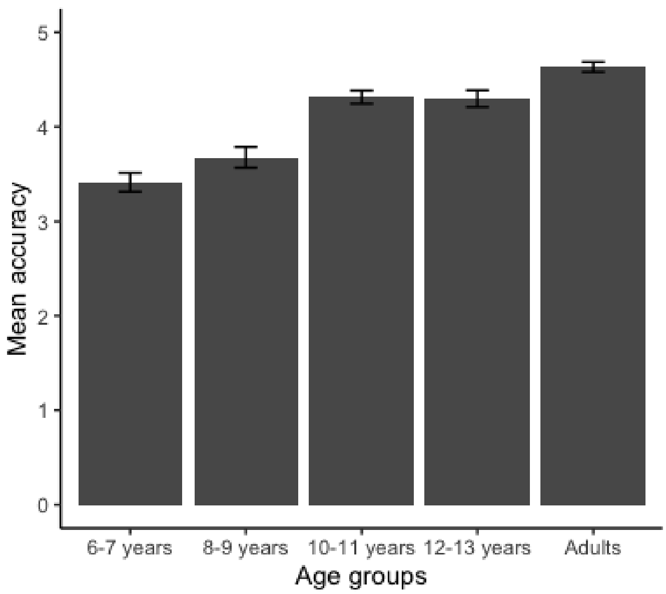

Figure 3: Mean values obtained for the accuracy of the hand laterality judgment task.
Groups 6-7 years and 8-9 years were less accurate than groups 10-11 years, 12-13 years, and adult (p <0.001). Error bars indicate standard error.

Reaction time

Figure 4 shows the mean RTs for the five age groups in the HLJ task. ANOVA identified a significant effect on RT ($F[4.180]=17.5$; $p<0.001$, $\eta^2=0.615$). Analysis with Bonferroni showed that the youngest group (6-7 years) was significantly slower than the other groups ($p<0.05$). Group 8-9 years was also slower than the groups 10-11 years, 12-13 years, and adult. We also found that the adult group did not differ regarding the RT when compared to the older children groups (groups 10-11 years and 12-13 years).

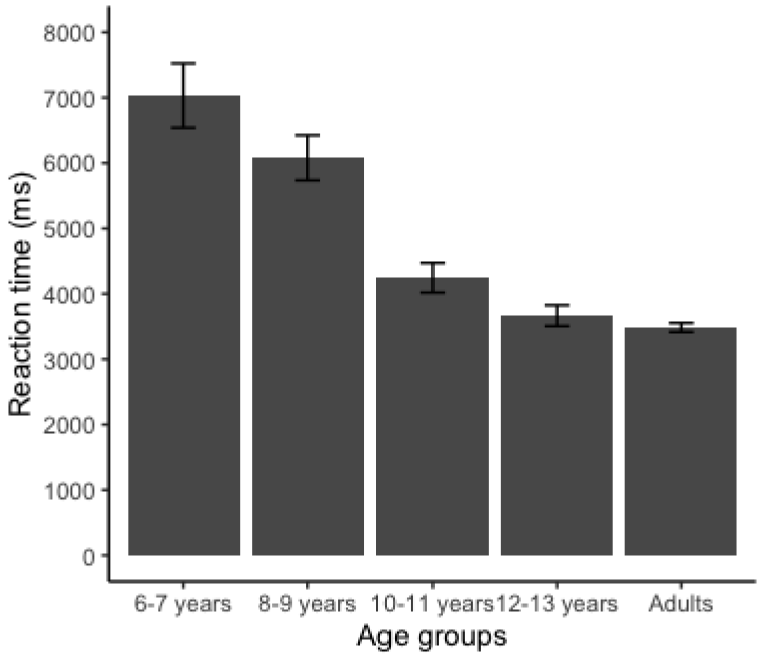

Figure 4: Reaction time (RT) for the hand laterality judgment task.
Group 6-7 years presented longer RT than groups 8-9 years, 10-11 years, 12-13 years, and adult ($p <0.001$). Group 8-9 years showed longer RT than groups 10-11 years, 12-13 years, and adult ($p <0.001$). Error bars indicate standard error.

Discussion

Our results revealed that the youngest children studied (group 6-7 years) were able to perform MI to solve the mental rotation task. There was a biomechanical restriction effect for all age groups, with all presenting lower RT to recognize the stimuli oriented in medial positions than the stimuli of lateral orientation. We also found that when task stimuli were presented in the dorsal view, the volunteers had lower RT to judge the stimuli. Finally, we observed a progressive improvement in the performance of the task as the age of the participants increased, reaching stabilization after 10 years, when the performance in the HLJ task was similar to that of the adult group.

The age at which children begin performing HLJ tasks using MI is not sufficiently clear in the literature. This is partly because the HLJ task is cognitively complex as it depends on the ability to mentally rotate images, on the ability to discriminate right and left, and on the ability to integrate visual and proprioceptive afferences. Several studies suggested that children may perform mental rotations at 5 years of age, albeit at a slower rate than adults (Frick, Hansen, & Newcombe, 2013; Funk, Brugger, & Wilkening, 2005; Marmor, 1977). According to Belmont and Birch (1963), it is expected that from the age of 6 the child will be able to recognize in himself / her right and left limb. Between 5 and 7 years old children acquire the ability to integrate visual and proprioceptive afferences necessary for the execution of movement (Guilbert, Jouen, & Molina, 2018; King, Oliveira, Contreras-Vidal, & Clark, 2012; Caeyenberghs et al., 2009). We found that the youngest children studied herein (group 6-7 years) used MI to solve the mental rotation task, suggesting that at these young ages children already have the cognitive requirements to perform the HLJ task. As our study did not involve children younger than six, the minimum age at which the ability to use MI to solve mental rotation tasks occurs remain an open question. Notwithstanding, our results indicate that because children 6-7 years old are able to use the mental rotation strategy, it is plausible to think that MI-based interventions could be used in this age group. This suggestion is supported by the literature review conducted by Spruijt et al., (2015). After analyzing some studies, Spruijt et al., (2015) suggest that MI training is a potential and viable method for the rehabilitation of children aged 5 years and older. Some studies involving populations aged 7 to 12 years highlight the potential of MI training in children (Souto et al., 2020; Doussoulin & Rehbein; 2011).

The effect of biomechanical restrictions on medial and lateral rotations was observed in all age groups. However, the accuracy is significantly reduced in the groups 6-7 years and 8-9 years, and the RTs of these children are higher than those presented by older children and the adult group. Our findings contrast those reported by Spruijt et al. (2015) because they found that 6 years old children were not able do not perform MI tasks. This divergence may be due to experimental approaches as these authors measured the timing of the actions imagined and performed, and not the HLJ task used herein. These contrasting results suggest that performance in MI may be task dependent. For Spruijt et al. (2015) the mental chronometry paradigm seems to be a conservative measure that may underestimate individuals' ability to use MI. In this study the authors found that not all healthy adult individuals used MI to solve the task. Thus, we believe that when considering the use of MI in pediatric rehabilitation, it is important for the child to make an individualized assessment of MI ability in order to ensure the effectiveness of the technique. Given the divergent results of studies using different tasks, it may be advisable to use multiple tasks to draw more definitive conclusions about children's ability to use MI.

The classic mental rotation task employed in our study has been widely used to evaluate MI (Butson et al., 2014; Conson, Elisabetta, & Luigi, 2013; de Lange, Helmich, & Toni, 2006). In this task the individuals are required to imagine their hand moving to judge the laterality of the stimulus, thereby making the task highly effective to assess motor information during the mental transformation of hand stimuli (Conson, Elisabetta, & Luigi, 2013). This is based on the hypothesis that the effect of biomechanical constraints is indicative of the use of the mental rotation strategy. Thus, the easiest physically executed stimuli are also judged faster supporting the idea that the same biomechanical factors that constrain actual movements also determine imagined movements (Williams et al., 2012). For Parsons (1994), presence of biomechanical effects provides clear evidence that MI has been used to solve the HLJ task.

Additional evidence for the use of the mental rotation strategy comes from the effects of the posture in which the hand was presented. Participants in our study recognized faster stimuli presented in dorsal view. Similarly, Butson et al. (2014) reported that children from 5 to 12 years old also presented lower RTs for dorsal view stimuli. Knowing that, to judge stimuli, individuals imagine their hand moving from the current position, a possible explanation for this finding would be that individuals remain with their hands in the dorsal posture while performing the task. Strengthening this hypothesis, previous studies suggested that the time to judge hand laterality is strongly influenced by the member's actual position during task resolution (Ter Horst, Van Lier, & Steenbergen, 2010; Parsons, 1994). Therefore, in judging the laterality of hand figures, volunteers simulate the movement of their own body from its present (egocentric) position, rather than from an allocentric representation. Shenton, Schwoebel and Coslett (2004) evaluated the influence of hand posture on the HLJ task, performing two judgment blocks: one with hands in dorsal posture and a second with hands in palmar posture. There were no significant differences in RT to judge the stimuli, indicating that hand posture during task resolution influences the RT spent to judge the stimuli. These observations suggest that, by recognizing still images of hands in varying positions, subjects move their own hands to their respective positions to arrive at a laterality decision.

In our study, only children from 10 years of age had the facilitating effect of dorsal vision to solve the HLJ task. One possibility is that the recognition of stimuli in dorsal vision represents a maturational effect on the HLJ task. Individuals tend to judge hand stimuli from their current position rather than from a fixed representation in the brain. We believe that the absence of this effect in younger children is due to the fact that, at this age, children did not go through the complete maturation of motor and cognitive processes involved in MI. According to Casey, Tottenham, Liston and Durston (2005), children show increasingly specialized motor and perceptual behavior. This is due to the fact that neural networks become increasingly differentiated with development. For these authors, these changes allow older children to process information faster and more accurately than younger children.

The effect of the presented hand posture is modulated by age. Groups involving children aged 10 and older find easier to judge laterality from the dorsal view. A possible explanation for this interaction may be the effect of visual influences. If the mental rotation strategy is used to decide on laterality from an egocentric perspective, the dorsal view is privileged. This effect may take a few years to develop depending on the individuals' experience. This interpretation is supported by evidence indicating visual influences on body schema as shown in the rubber hand experiment (Shenton, Schwoebel, & Coslett, 2004).

We hypothesized that there would be changes in MI ability as age increased. Our results support this hypothesis by showing progressive improvement in the performance of the HLJ task as the participants' age increased. It is important to highlight, however, that the improvement in motor imaging performance occurred in children up to 10 years old. From that age, performance was similar to that of adults. In line with our results, most studies using the HLJ paradigm also reported increased motor involvement with age (Spruijt, van der Kamp, & Steenbergen, 2015; Conson, Elisabetta, & Luigi, 2013; Toussaint et al., 2013; Caeyenberghs et al., 2009). The study by Caeyenberghs et al. (2009) compared the performance in MI through the HLJ task of 7 and 8 year olds, 9 and 10 years old and 11 and 12 years old. The results showed that older children were faster and more accurate than younger children, suggesting changes in MI as they age. Strengthening this hypothesis, the articles on age-related differences in MI analyzed in the Spruijt et al., (2015) review indicate that children's ability to perform the task accurately increases with age.

Indeed, from 10 years old, the performance in the HLJ task resembled that of the adult group. We also found a progressive decrease in RT as participants' age range increased. Children of 6-7 years old were slower than those of the other age groups and children aged 8-9 years were also slower when compared to older age groups. Indeed, the performance in the HLJ task of children aged 10 and older was similar to that of adults. We found that the adult performance level with regards to accuracy and RTs is reached when children reached 10 years of age. This result probably reflects the maturation of the brain areas (posterior parietal cortex, premotor area, cerebellum and frontoparietal region) involved with the mental simulation of body part movements (Butson et al., 2014; Caeyenberghs et al., 2009; Choudhury, Charman, Bird, & Blakemore, 2007).

Our results point to an improvement in MI capacity as age increases. Similar results were also found by Caeyenberghs et al. (2009). This improvement in MI as age is supported by the development and maturation of a set of complex cognitive processes (Caeyenberghs et al., 2009). Significant structural and functional changes occur in the child's brain during childhood. According to Casey et al. (2005) children show increasingly specialized motor and perceptual behavior due to the fact that neural networks become increasingly differentiated with development. For these authors, these changes allow older children to process information faster and more accurately than younger children. Casey et al. (2005) further state that fronto-parietal coupling is greatly increased throughout childhood, in particular between 6 and 10 years of age. This explains why the children in our study showed progressive improvements in performance with age, as well as a similar response pattern to adults when they reached the age of 10 years.

Our results point to a non-linear improvement in RT, corroborating the findings of Fuelscher et al., (2016). We found that the ability of MI progressively improves until 10 years of age, after that age, the performance is similar to that observed in adults. Thus, as in previous studies (Fuelscher et al., 2016; Caeyenberghs et al., 2009), our study points to a substantial maturation in MI ability in the early years of elementary school, becoming mature in late childhood and early adolescence.

For Fuelscher et al., (2016) there is evidence that the development of MI can also be influenced by the development of general cognitive factors, such as the visuospatial capacity of working memory. Indeed, these interindividual differences in MI ability can be explained by cognitive and motor skills that may facilitate or restrict the development of MI. Previous studies suggest that executive functioning, planning ability, movement experience, working memory, and intelligence may all influence MI (Spruijt, van der Kamp, & Steenbergen, 2015; Caeyenberghs et al., 2009; Krüger M, Krist, 2009; Malouin, Belleville, Richards, & Desrosiers, 2004). Nonetheless, we suggest that MI is a continuous and progressive refinement throughout childhood and early adolescence, becoming progressively stronger with advancing age. We attribute the maturation in MI capacity to the development of neural networks linked to the internal simulation of movements. This maturation in the ability to perform imagined movements can be interpreted in terms of a general development of the cognitive processes involved in motor representation. This development is mainly determined by internal changes in the structures of the prefrontal and parietal cortex (Sirigu & Duhamel, 2001). This is in line with previous evidence that the parietal cortex is involved in the formulation of internal models associated with motor imagery and the internal representation of action (Sirigu & Duhamel, 2001). Vargas et al. (2004) also point out that the evolution of MI in children is also related to the maturation of the supplementary motor area, premotor area, primary motor cortex, basal ganglia and cerebellum.

Limitations and Implications

Our results provided evidence that children aged 6 years and older are able to use MI to solve the mental rotation task. However, as our study did not involve children under 6 years old, the minimum age at which this ability is present remains an open question, which is a limitation of this study. With a sample composed of ages ranging from 6 to 13 years, our results suggest that there is a progressive improvement in MI as age increases. These results are in line with previous studies (1, 20, 21, 39). However, it is not yet possible to make definitive inferences about the exact trajectory of development. For this, studies with longitudinal methodological design would be necessary.

Due to the characteristics of the MI skill, we believe the divergent results are due in part to the use of different tasks. In addition, individual differences may also influence this ability, such as cognitive functioning. Studies suggest that working memory, attention, planning, and intelligence may facilitate or restrict the development of MI (Spruijt, van der Kamp, & Steenbergen, 2015; Caeyenberghs et al., 2009; Krüger & Krist, 2009; Malouin et al., 2004). According to previous studies, motor planning ability and motor skills may also influence MI performance (Barhoun et al., 2019; Fuelscher et al., 2016). However, our methodological design did not include measures to assess these skills, which is one of the limitations of the present study. Thus, experiments that evaluate the development of MI controlling cognitive and motor skills are still a challenge for future studies.

The use of motor imagery by children has important theoretical implications. Recent studies suggest that performing MI activates specific sensorimotor representations involved in the planning and execution of motor acts (Steenbergen, Crajé, Nilsen, & Gordon, 2009). Thus, MI is a useful tool in pediatric rehabilitation. Few studies have investigated the use of MI in the rehabilitation of children. Buccino et al. (2012) applied MI training by observing action associated with real movements in children with cerebral palsy and found beneficial results. In this experiment, the authors observed that the group of children who watched other people's videos producing actions led to an increase in motor function, which was not observed in children who watched videos without motor content. One advantage of the implicit use of MI by observing the action is that it can be beneficial for small children who cannot be educated on the use of MI. Our results provide contributions about the development of MI in children setting an important starting point for future research interested in assessing the effectiveness of MI as a tool for pediatric rehabilitation.

Conclusion

The use of the mental rotation strategy by 6-7 year-olds has important theoretical implications and further investigation of the neuro-cognitive foundations is warranted. The results obtained herein indicating the influence of biomechanical restrictions and hand posture suggest that children use the strategy of mental representation of the body part. Future research needs to clarify the role played by hand laterality judgment, mental object rotation, and cognitive control processes in HLJ execution. Our results also have important clinical applications. There is currently a strong interest in the use of MI-based interventions for the development and rehabilitation of cognitive and motor functions and the results presented herein indicate that this strategy may be used in children as young as six.

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STUDY 3: ABILITY OF CHILDREN WITH CEREBRAL PALSY TO USE MOTOR IMAGERY FOLLOWS MECHANICAL CONSTRAINTS AND DOES REQUIRE WORKING MEMORY

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Abstract

Aims: This study evaluated whether children with cerebral palsy are able to engage in a motor imagery task. Possible associations between motor imagery and functional performance, working memory, age and intelligence were also investigated. Methods: The study assessed 57 children with unilateral cerebral palsy (UCP) [mean age = 10 years and 4 months (sd = 2.1 years)] and 175 healthy (control) children [mean age = 9 years and 4 months (sd = 1.95 years)]. The hand laterality judgment (HLJ) task was used to measure motor imagery ability. Reaction time, accuracy, and the effect of the biomechanical constraints were assessed in this task. Results: Performance in both groups followed the biomechanical constraints of the task. Reaction time means did not differ significantly between groups ($p > 0.05$). Significant differences between the UCP and control groups were observed for accuracy ($p < 0.05$). Functional performance and working memory were correlates of motor imagery tasks. Interpretation: Results suggest that children with UCP can engage in motor imagery, however, they commit more errors than healthy controls. In addition, their performance in tasks of motor imagery is influenced by functional performance and working memory.

Keywords: motor imagery; unilateral cerebral palsy; hand laterality judgment task, functional performance; working memory.

Introduction

Individuals exhibiting unilateral cerebral palsy (UCP) present various motor impairments resulting from unilateral injury to their immature brain (Bax et al., 2005). There is evidence that motor deficits in individuals with UCP are caused by difficulties in motor performance and planning (Craje et al., 2010; Mutsaarts, Steenbergen, & Bekkering, 2006). Steenbergen, Meulenbroek, and Rosenbaum (2004) proposed that these planning deficits may be more prominent in patients with right hemiplegia, as motor control relies heavily on the left brain hemisphere. However, the injury resulting from the UCP can lead to cortical reorganization and, in some cases, the affected upper extremity becomes more controlled by the ipsilateral corticospinal tract (Kuo et al., 2017).

It has been proposed that motor imagery (MI), i.e., imagined movements, critically contributes to the movement planning process and is a prerequisite for motor planning (Jeannerod, 1994). A growing number of studies using functional magnetic resonance imaging have shown similar cortical activation patterns during physically implemented and imagined movements (Kuoet al., 2017; Grezes & Decety, 2001).

According to Mutsaarts, Steenbergen, and Bekkering (2006), motor planning deficits observed in UCP may be related to a reduced ability to use MI. To investigate this hypothesis, several studies tested the MI ability of children and adolescents with UCP (Williams, Anderson, Reid, & Reddihough, 2012; Craje et al., 2010; Steenbergen, van Nimwegen, & Crajé, 2007; Mutsaarts, Steenbergen, & Bekkering, 2006) using variations of the mental rotation tasks, such as the hand laterality judgment – HLJ (Parsons, 1994). In this task, hand figures are presented in different spatial orientations (eg.: 0º, 90º, 180º e 270º), and individuals mentally simulate the movements of their own hands and then judge the laterality of the stimuli. The use of MI as a strategy is indicated when the biomechanical s that affect real movements, also affect the imagined task (Conson, Mazzarella, & Trojano, 2013; Parsons, 1994). The effect of biomechanical constraints refers to the increase in RTs when hand figures are presented in anatomical positions that make mental rotation difficult (Figures with fingers facing sideways). Similarly, a decrease in response time is observed when the stimuli are medially rotated (figures with fingers facing medially).

Studies that applied the HLJ task to children and adults with typical development reported that the reaction time (RT) of medially oriented stimuli (see Figure 1) is biomechanically easier to perform, resulting in shorter RTs compared with stimuli oriented laterally (Souto et al., 2020). Results of studies using the HLJ task to study MI ability in individuals with UCP are heterogeneous. Mutsaarts, Steenbergen, and Bekkering (2007) reported that only individuals with right UCP could not perform MI. In contrast, Jongsma et al. (2016) described the presence of MI ability in UCP only when the unaffected side is involved. However, Steenbergen, van Nimwegen, and Crajé (2007) found that both right and left hemiplegics could perform the HLJ task, albeit less accurate and slower than controls. Finally, Williams et al. (2001) observed that children with UCP can perform MI tasks, regardless of the impaired side. A possible explanation for these inconsistencies is the limited sample size used in these studies. In addition, another factor that could explain the divergent results is the lack of control of some variables that can influence the MI ability, such as functional performance and working memory.

MI ability may depend on individual functional performance. The results of Williams et al. (2012) suggest that poor motor skills lead to reduced MI ability. However, these results should be interpreted with caution because the instrument used to evaluate the functional performance was a subjective questionnaire applied to the children's parents. A second factor that may limit success in MI tasks is working memory. During the imagery process, information is retained and processed in working memory (Malouin et al., 2004). Thus, working memory deficits could impair the effectiveness of MI tasks. The possible association between MI and working memory performance in UCP has not been explored in the existing literature.

Together, the studies published to date do not allow for a definitive conclusion on the MI ability of individuals with UCP. Knowing that imagined movements are important in the planning and execution of movements, it becomes relevant to determine whether UCP individuals can perform MI tasks. Furthermore, verifying MI integrity in this population constitutes an important step for future experiments investigating MI as a tool in motor rehabilitation. The HLJ task was applied to 57 UCP children with lesions in the right or left brain hemispheres without intellectual disability. The performance of these children, which present several functional performance and working memory levels, was compared to that of 175 healthy children.

The HLJ task was used to test the following hypotheses: 1) Children with UCP and healthy individuals should use similar MI strategies; therefore, performance in the HLJ task should obey the biomechanical constraints (increase in RT as a function of the rotation angles) for both groups. 2) Given that MI plays an important role in motor planning (Craje et al., 2010; Steenbergen, van Nimwegen, & Crajé, 2007) and knowing that motor planning is compromised in the UCP (Craje et al., 2010; Mutsaarts, Steenbergen, & Bekkering, 2006), a worse performance in MI (longer RTs and less accuracy) should be observed in these individuals. 3) Since ipsilateral motor tract reorganizes (Kuo, Friel, & Gordon, 2018), differences in RT and accuracy should not be found between children with right and left UCP. 4) Functional performance scores of children with UCP should correlate with higher accuracy rates and shorter RT. 5) Individuals with UCP who have better working memory scores should have better accuracy rates and lower RT than those with lower scores.

Methods

Design and participants

This is a cross-sectional study in which the sample was obtained for convenience. The study included 57 UCP children [mean age = 10 years and 4 months (SD=2.67 year)] recruited in non-governmental organizations that provide rehabilitation services for the unprivileged. The participants were divided into two groups: RUCP, composed of 32 volunteers [mean age = 10 years and 2 months (SD=2.88 years)] and LUCP with 25 volunteers [mean age = 10 years and 5 months (SD=2.44 years)]. The control group comprised 175 children with no history of neurosensory and neuropsychiatric impairments [mean age = 9 years and 4 months (SD=2.14 years)]. Volunteers with a diagnosis of UCP and healthy controls aged 6 to 14 years were included. All children had normal or corrected vision and the ability to discriminate between right and left. No individuals with UCP presented intellectual disability or deficit in working memory. Controls were recruited at a state-run school. All included volunteers came from a similar socio-economic and cultural background, as they all attended free state-run schools and non-governmental rehabilitation centers.

Ethics

All research procedures were approved by the Research Ethics Committee of the Federal University of Minas Gerais (COEP / UFMG). The consent was obtained in writing from the parent/guardian of all children prior to their assessment. Participation was dependent on the children's oral consent.

Assessment measurements

Intelligence

Intelligence assessment was carried out to ensure that there would be no systematic differences in general intelligence between participants with left and right UCP. We also examined possible correlations between IQ scores and performance in the HLJ task (accuracy and RT) in children with UCP. Initially, the age group was defined between 6 to 11 years, and the Raven's Coloured Progressive Matrices test was used (Angelini,et al., 1999). However, due to the small study sample obtained, a new age group was established, including volunteers up to 14 years old. For adolescents aged 12 to 14 years, the Wechsler Intelligence Scale for Children Fourth Edition - WISC IV Vocabulary and Block Design subtests were used (Wechsler, 2002). The scores of the two intelligence tests were transformed into a z score to obtain a single measurement. There were no a priori cutoff points for intelligence.

Right–left orientation

The Right–Left Orientation Test was used (Dellatolas, Viguier, Deloche, Agostini, 1998). The test has 12 items of right and left body parts recognition. It is divided into three steps: the first presents simple commands regarding the child's own body; the second consists of double commands – direct and crossed – toward the child's body; and the third commands the child to point to single lateral body parts of an opposite-facing person. Correct answers were scored as one and wrong answers scored as zero. All participants exhibited 80% or more accuracy in the Right-Left Orientation Test.

Working memory

The backward Corsi Cubes test (Santos, Mello, Bueno, & Dellatolas, 2005) was used to assess working memory in children with UCP (Table 1). This test involves a wooden board in which 9 blocks of the same dimensions are irregularly distributed. The blocks are identified only by the examiner by the numbers on their faces. During the test, the examiner first touches a series of blocks with the index finger, then the participant must point the blocks in the backward order in which the examiner pointed them. The number of blocks is progressively increased and testing is interrupted when the individual makes two consecutive errors. Spatial memory capacity is defined by the most extensive series correctly recalled (Santos et al., 2005). An a priori cutoff was not established because we aimed to assess its relationship with the MI task.

Functional performance.

Assisting Hand Assessment – AHA (version 4.3) is an instrument used to assess the efficiency with which a child with unilateral motor disability uses the affected upper limb during activities that require bimanual coordination (Krumlinde-Sundholm, Holmefur, Kottorp, & Eliasson, 2007). First, a 10-15 minute session is recorded on video with a specific toy from the AHA test kit, which requires bimanual manipulation. The video recordings are subsequently analyzed, considering 22 predefined items, using a rating scale ranging 1 to 4 points. The sum of the gross score ranges from 22 (low capacity) to 88 points (high capacity). The instrument has excellent reliability and validity. Considering that the objective was to establish the functional performance of the hands, no cutoff points were established for AHA.

Motor imagery task

A modified version of the HLJ task was used to assess MI ability (Parsons, 1994). In this task, drawings of the back and palm views of a young (female) adult's hands were presented individually in the center of the screen of a laptop (13 inches). The HLJ task was programmed with the Presentation software, version 0.71. The hands were presented pseudorandomly at different rotation angles (0º, 90º, 180º, and 270º) and remained on the screen until the individual registered his/her response by pressing a designated key on the laptop keyboard (Figure 1). This version of the task consists of 16 different hand positions, repeated five times each, totaling 80 stimuli. The participants remained seated in a comfortable position and were instructed to decide, as quickly and precisely as possible, if each stimulus was a left or right hand. All participants were instructed to remain with the palm facing downwards and to not move their hands or heads during the task. Participants with UCP were instructed to respond using the index and middle fingers of the non-affected limb. Participants had to press the right button when the stimulus was a picture of a right hand and the left button for a left hand picture. The computer keys were marked with stickers to label them as left and right. Prior to the experiment, eight figures of the hands at different angles of rotation were shown to the participants for initial practice training. The actual experiment lasted approximately 15 minutes.

For each stimulus, RT and accuracy were recorded. RT data allowed to analyses the effect of biomechanicals to confirm the use of MI by the participants. This effect is characterized by changes in RT, as a function of the stimulus rotation angle. Thus, medially rotated hand figures are easier to recognize than laterally oriented figures (Conson, Mazzarella, & Trojano, 2013; Parsons, 1994). Therefore, the RT obtained to recognize stimuli in medial orientations (right hand 270º, left hand 90º) is lower than the RT obtained to recognize stimuli in lateral orientations (right hand 90º, left hand 270º). This effect is present when the biomechanical characteristics that affect physical movements also influence the imagined movement, thus confirming the use of MI (Parsons, 1994). RT and accuracy were the dependent measures used to assess the effect of biomechanical constraints.

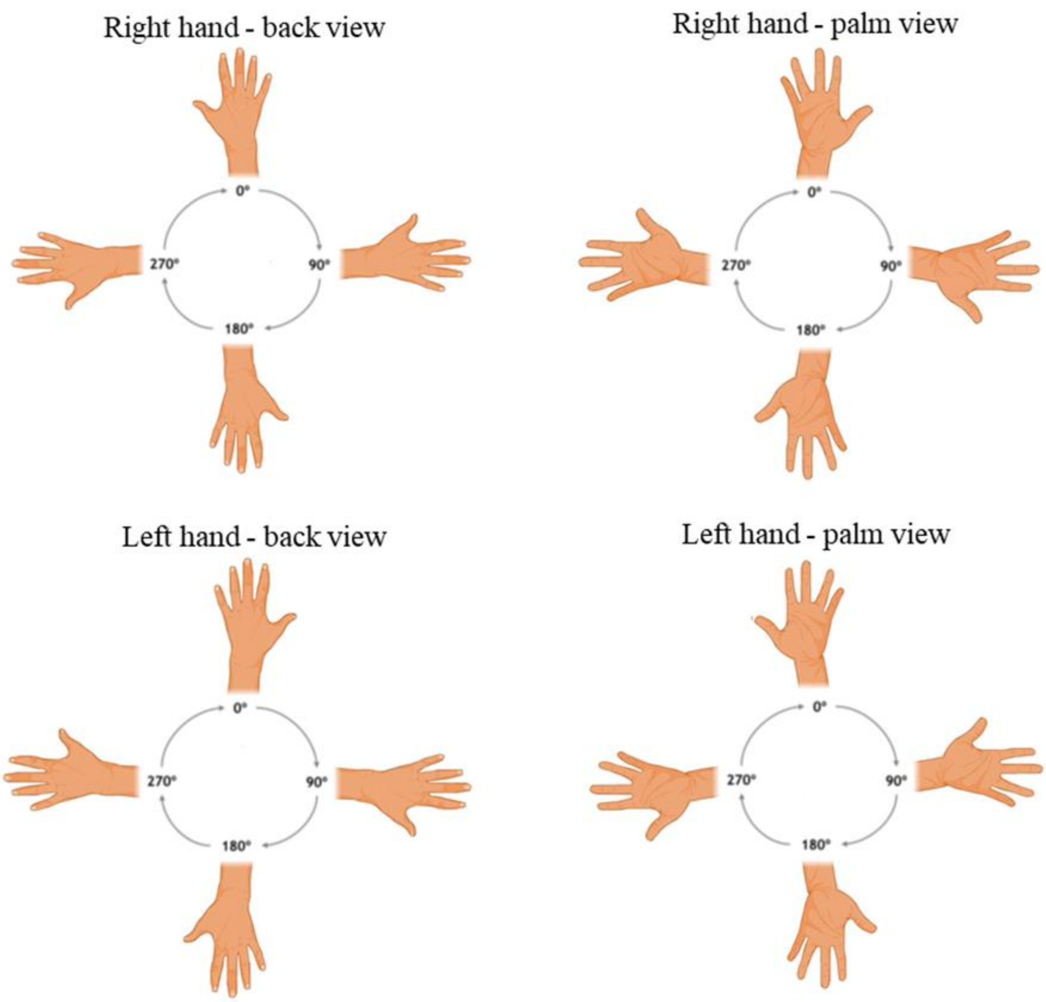

Figure 1: Stimuli for the task of judging hand laterality. The stimuli of the right hand oriented at 270º and left hand oriented at 90º correspond to medial rotation. The stimuli of the right hand oriented at 90º and the left hand oriented at 270º correspond to lateral rotation.

Statistical analyses

Analyses were performed using SPSS (version 20.0). The level of significance was defined at $p < 0.05$. The sample's homogeneity in relation to sex was assessed with the chi-square test and, in relation to age, with one-way ANOVA followed by post hoc Bonferroni tests. The Student's t test for independent samples was used to compare intelligence, functional performance, and working memory between the UCP groups. For the HLJ task, stimuli with incorrect answers or RT greater than three standard deviations above or below the subjects' grand mean were excluded from the analyses. These procedures were performed to control for possible impulsivity or distraction effects. The paired-sample t-test was used to evaluate biomechanical constraints, as performed by Crajé et al. (2010). To this end we compared the general average RT obtained for the medial (270º of the right hand and 90º of the left hand) and lateral (90º of the right hand and 270º of the left hand) rotations. Cohen's d was used to verify the magnitude of the biomechanical constraints effects.

Repeated measures ANOVAs were used for analyses of RT and accuracy. Three within factors were included in ANOVA: rotation angle (4 angles: 0º, 90º, 180º, 270º), view of the stimulus (2: back and palm view); hand (2: right and left hand). One between-groups factor was used: group (control, RUCP, LUCP). The multivariate approach was utilized to protect against violations of the assumption of sphericity. Post hoc tests were conducted using the Bonferroni procedure and the partial eta-squared (η^2) was calculated to determine effect size.

Finally, multiple linear regression was performed using the general RT and accuracy averages obtained from the UCP groups as a dependent variable. The following independent variables were included in the regression model: age, intelligence, functional performance, and working memory.

Results

The three groups were homogeneous in relation to sex (p=0.652) but not in relation to age (p=0.016), with the controls being significantly younger than LUCP (p=0.047). The groups RUCP and LUCP did not differ significantly in relation to age (p=1.000). The comparison between the groups for intelligence, working memory, and functional performance are shown in Table 1.

Table 1. Group means (SD) for descriptive measures.

	Controls (n= 175)	RUCP (n= 32)	LUCP (n= 25)	Statistic tests	*p*-value
	Mean (SD)	Mean (SD)	Mean (SD)		
Age*	9.39 (2.14)	10.22 (2.72)	10.56 (2.36)	F(2.230)=4.236	0.016
Gender**					
Female	87	15	10	$\chi 2$=0.856	0.652
Male	88	17	15		
Intelligence***	-	-0.034 (0.586)	-0.119 (0.463)	*t (55)*=0.596	0.554
Working memory***	-	2.94 (1.366)	2.84 (0.987)	*t (55)*= 0.301	0.765
Functional performance***	-	60.06 (15.97)	60.16 (17.43)	*t (55)*=-0.022	0.983

Note: *ANOVA to compare 3 groups.
**Chi Square for cathegorical variable.
***Independent sample test.
Level of significance: *p*<0.05
Controls: children with typical development; LUCP: children with left unilateral cerebral palsy; RUCP: children with right unilateral cerebral palsy; SD: standard deviation

Specific results will be covered in the following sections: first, the analyses regarding the effects of the biomechanical constraints on the HLJ task and then the results of RT and accuracy will be presented. Finally, we investigate whether intelligence, age, functional performance, and working memory correlate with the performance (RT and accuracy) in the HLJ task.

Effect of biomechanical constraints.

The comparisons of the means of the RTs for the lateral and medial views are shown in Table 2. In this case, a paired t test was used. In the right hand, all three groups showed differences between medial and lateral rotations: Control [t(171)=5.79; p<0.001]; RUCP [t(31)=2.29; p<0.029]; LUCP [t(24)=2.17; p<0.040]. In the left hand the same results was found: Control [t(171)=5.49; p<0.001]; RUCP [t(31)=2.05; p=0.049]; LUCP [t(24)=4.61; p<0.001]. Thus, for both hands, lower RTs were observed when stimuli involving the medial rotation was compared with those for the lateral rotation.

Table 2: Effect of biomechanical constraints: Comparison between the means of lateral and medial rotations for each group.

Groups	Right hand						Left hand					
	Medial rotation	Lateral rotation	*t*	*df*	*p**	*d*	Medial rotation	Lateral rotation	*t*	*df*	*p**	*d*
	Mean (SD)	Mean (SD)					Mean (SD)	Mean (SD)				
Controls (n=175)	3620.61 (984.77)	4370.43 (1522.48)	5.79	171	0.001	0.56	3629.65 (1131.04)	4242.54 (1249.17)	5.49	171	0.001	0.50
RUCP (n=32)	3703.33 (663.71)	4429.59 (1612.61)	2.29	31	0.029	0.57	3796.39 (494.09)	4149.66 (831.03)	2.05	31	0.049	0.50
LUCP (n=25)	3899.64 (963.05)	4657.82 (1278.64)	2.17	24	0.040	0.65	3805.18 (592.12)	4636.32 (847.61)	4.61	24	0.001	0.99

* paired *t* test

Level of significance: $p<0.05$

Note: Controls: children with typical development; LUCP: children with left unilateral cerebral palsy; RUCP: children with right unilateral cerebral palsy; SD: Standard Deviation

Reaction time

Repeated-measures ANOVA showed an effect of the rotation angle on the RT [F(1.226)=62.9; $p<0.001$; $\eta^2=0.22$]. However, a significant main group effect was not found (F[2.226])=1.453; $p<0.103$; $\eta^2=0.07$) suggesting that the linear relationship between rotation angle and RT was present in the three groups. Furthermore, no significant differences were found in the stimuli representing the right or left hand (F[2.226])=2.65; p=0.104; $\eta^2=0.012$). There is no interaction between the hand side and the view (F[2.226])=1.371; p=0.243; $\eta^2=0.06$); between angle, hand and view (F[2.226])=2.363; p=0.070; $\eta^2=0.10$); and between rotation angle and group (F[2.226])=1.468; p=0.103; $\eta^2=0.08$). Significant differences were found to recognize the palm or back view hand stimuli (F[2.226])=4.096; p=0.044; $\eta^2=0.018$). There is an interaction between angle and hand (F[2,226])=11.860;p<0.001;$\eta^2=0.050$) which prevents the analyses of these two factors separately. Post hoc analyses, with Bonferroni correction for the back and palm views, revealed a difference between the views at 180º (p=0.005) and 270º (p=0.040) angles. At the 180º angle, the palm view showed a longer RT than the back view. For the 270º angle, the back view presented a longer RT than the palm view. The values of the means and standard deviations for the RT and accuracy are shown in Table 3.

Accuracy

Repeated-measures ANOVA for accuracy revealed a significant difference for rotation angle (F[1.226]=60.956; $p<0.001$; $\eta^2=0.254$). In addition, a significant main group effect was found (F[2,226])=27.59, p<.001; $\eta^2=0.198$). Post hoc analyses with Bonferroni procedure revealed that the controls were more accurate than both groups with UCP (p <0.001). No differences were found between participants with RUCP and LUCP (p>0.05). No significant differences were found for the right or left hand stimuli (F[2.226]=0.189; p=0.664; $\eta^2=0.001$) and for the back and palm views (F[2,226]=2.574; p=0.110; $\eta^2=0.015$). An interaction between rotation angle and view (F[2.226]=50.228; p<0.001; $\eta^2=0.224$) was observed. For the 90º and 270º angles, the back view was more accurate than the palm view (p=0.021 and p<0.001, respectively). For the 180º angle, the palm view was more accurate than the back view (p=0.001). There was no interaction between angle, hand, and view (p=0.083). The values of the means and standard deviations for RT and accuracy are shown in Table 3.

Table 3. Means and standard deviations of the RTs and accuracy in the HLJ task for the 3 groups.

Stimuli	Controls (n=175)		RHCP (n= 32)		LHCP (n=25)	
	RTs	Accuracy	RTs	Accuracy	RTs	Accuracy
	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)
0° (right hand, palm view)	3223.24 (1515.38)	4.37 (0.88)	3199.52 (864.99)	4.25 (0.71)	3406.56 (925.51)	3.92 (0.91)
90° (right hand, palm view)	4298.06 (1680.10)	3.23 (1.22)	4456.51 (1631.94)	2.88 (1.04)	4645.68 (1278.81)	2.92 (115)
180° (right hand, palm view)	4805.16 (2857.47)	3.92 (1.09)	4954.90 (1800.84)	3.31 (0.96)	5065.28 (1750.88)	3.46 (0.96)
270° (right hand, palm view)	3487.58 (1123.53)	4.14 (1.04)	3698.13 (1057.19)	3.41 (0.83)	3802.64 (813.53)	3.48 (1.04)
0° (left hand, palm view)	2732.57 (2060.58)	4.21 (0.97)	2843.13 (849.353)	3.97 (0.89)	3338.44 (867.22)	4.12 (0.95)
90° (left hand, palm view)	3621.52 (1436.59)	3.97 (1.28)	3797.42 (584.82)	3.49 (1.26)	3887.84 (687.84)	3.52 (1.63)
180° (left hand, palm view)	4804.29 (6295.25)	3.89 (1.39)	5108.65 (1185.58)	3.37 (0.84)	4936.34 (1537.47)	3.24 (0.97)
270° (left hand, palm view)	3998.18 (1603.42)	3.17 (1.37)	3994.04 (700.51)	2.89 (1.15)	4400.04 (895.54)	2.92 (0.99)
0° (right hand, back view)	3155.62 (2372.31)	4.38 (0.81)	3277.16 (1140.17)	4.08 (1.24)	3012.92 (1116.42)	4.04 (1.17)
90° (right hand, back view)	4393.07 (1738.18)	3.53 (1.44)	4398.52 (1631.94)	2.94 (1.55)	4669.96 (1279.51)	2.98 (1.12)
180° (right hand, back view)	4237.53 (1927.96)	3.59 (1.37)	4330.55 (2160.51)	3.29 (1.17)	4239.07 (1967.97)	2.88 (0.97)
270° (right hand, back view)	3770.11 (1374.74)	4.20 (1.30)	3743.48 (712.06)	3.09 (1.11)	3996.64 (1427.04)	3.26 (1.18)
0° (left hand, back view)	3113.83 (1308.51)	4.44 (0.95)	3519.35 (1220.40)	4.03 (0.97)	3511.24 (1185.69)	4.07 (1.15)
90° (left hand, back view)	3604.20 (1441.89)	4.15 (1.25)	3774.52 (740.94)	3.72 (0.97)	3822.09 (650.41)	3.92 (1.17)
180° (left hand, back view)	4362.59 (1954.08)	3.88 (1.38)	4532.59 (2098.97)	3.25 (1.12)	4563.16 (1916.05)	3.16 (1.49)
270° (left hand, back view)	4490.30 (1491.88)	3.69 (1.51)	4305.23 (1277.38)	2.85 (1.13)	4872.61 (964.29)	2.88 (1.81)

Note: Controls: children with typical development; LUCP: children with left unilateral cerebral palsy; RUCP: children with right unilateral cerebral palsy; RT: Reaction times; SD: standard deviation.

Functional performance and working memory predicts performance on the HLJ task

Multiple linear regression was used to verify whether functional performance, working memory, intelligence, and age would be associated with the performance in the HLJ (RT and accuracy). The analyses for RT resulted in a significant model [F(1.55)=23.761; p<0.001; R2=0.468]. Functional performance and working memory are predictors of HLJ task performance; however, age and intelligence are not significant (p>0.05). The analyses for accuracy also resulted in a significant model [F(1.55) = 16.976; p<0.001; R2=0.236]. Functional performance and working memory are significant predictor of HLJ task performance but age and intelligence are not significant (p>0.05). Regression analyses are shown in Table 4.

Table 4. Regression analysis for RT and Accuracy.

Predictor	RT (adjusted R^2 = 0.448)				Accuracy (adjusted R^2 = 0.236)			
	Beta	Partial t	Significance	r^2 change	Beta	Partial t	Significance	r^2 change
Intercept		22.137	<0.001			12.012	<0.001	
Age	0.031	0.056	0.758	Excluded	0.030	0.252	0.802	Excluded
Intelligence	0.017	0.174	0.867	Excluded	0.159	1.322	0.192	Excluded
Functional performance	-0.534	-4,697	<0.001	0.468	0.493	4.453	<0.001	0.236
Working memory	-0.241	-2.121	<0.001	0.468	0.201	3.120	<0.001	0.236

Note: RT: Reaction Time. Level of significance: p<0.05

Discussion

Performance of children with UCP was compared to that of healthy children on the HLJ task, a widely used task to assess MI ability, to verify if their performance followed the biomechanical constraints indicative of MI use. Additionally, the association among HLJ, functional performance, and working memory were investigated in children with UCP. It was hypothesized that if children with UCP and healthy individuals use similar MI strategies, their performance in the HLJ task should obey the task's characteristic biomechanical constraints. Both UCP groups and controls responded faster to medially than laterally oriented hands, suggesting that they followed biomechanical constraints and used similar MI strategies to solve the HLJ task. This result is consistent with a previous study using HLJ variations to evaluate the presence of MI abilities in individuals with UCP (Williams et al., 2011). This result is also consistent with Fitts' law (1954), which shapes the speed-accuracy relationship where the task's difficulty is associated with increasing execution times. Thus, the effect of biomechanical constraints is compatible with the Fitts law. The biomechanical constraints effect is present when the characteristics that affect real movements also influence the imagined task. This occurs because the easiest physically executed stimuli are also judged faster (Parsons, 1994). Butson et al. (2014) suggested that children with typical development use the egocentric perspective to solve the HLJ because their performance is affected by biomechanical constraints, that is, they need to imagine their hands rotating toward the position of the displayed stimulus to complete the task. Thus, we suggest that the children with UCP also solved the HLJ task from an egocentric perspective.

It was hypothesized that worse performance in MI (longer RTs and less accuracy) should be observed in children with UCP. As for RT, this hypothesis cannot be firmly supported. Although children with UCP presented slower responses than controls, the differences were not significant. These findings are consistent with the work of Williams et al. (2011). However, they are at odds with the results found by Steenbergen, Van Nimwegen and Crajé (2007), who found that the RT of children with UCP was significantly slower than that of the controls. These authors explained the low RT performance of participants with UCP by the strategy employed in solving the task suggesting that children with UCP used visual instead of motor imagery as an alternative to solve the HLJ task. They also proposed that participants with UCP consider the stimuli as figures, and not as parts of the body, thus suggesting that the children perform mental rotation from a third-person’s perspective. However, if the participants in our study used visual imagery, there would be no changes in the RT as a function of the rotation angle, and consequently, the effect of biomechanical constraints would not be present. Researchers claim that the effect of biomechanical constraints is unique to MI and should not be present when individuals use visual imagery to solve the HLJ task (Butson, et al., 2014; Lust, Geuze, Wijers, & Wilson, 2006). Furthermore, during the HLJ task, the children maintained their hands in the back view; thus, if the children were involved in visual imagery, there would be longer RTs to recognize the palm view stimuli, which the child cannot see during the task. The present study found no significant differences between back and palm views, with the exception of the 180º and 270º angles that were longer in the palm and back views, respectively, suggesting that children were not involved in visual imagery. One way to avoid using visual imagery to solve the HLJ task is to cover the child's hands with a cloth (Steenbergen, van Nimwegen, & Crajé, 2007) however, this procedure was not performed in our study.

Taking together, we suggest that participants with UCP involved MI to solve the HLJ task, despite the slightly no significant slower RT. It is possible that, despite motor impairment, children with UCP activate the neural networks involved in the imagined movement, as suggested by functional neuroimaging studies Chinier et al. (2014). Indeed, Chinier et al. (2014) showed activation of the bilateral fronto-parietal network of individuals with UCP during the execution of MI tasks, which are the same neural substrates involved in MI in healthy individuals (Hétu et al., 2013).

The results of the present study are intriguing since the groups with UCP showed the effect of the biomechanical constraints and thus did not differ from the controls for RTs; however, their accuracy in solving the HLJ did not match that of healthy children. Steenbergen, van Nimwegen, & Crajé (2007) also used the HLJ task in UCP children and found similar results regarding accuracy and RT, although they did not examine the role of biomechanical constraints. These findings indicate that UCP groups can perform MI tasks, but their ability does not match that of healthy children.

It was hypothesized that there would be differences in RT and accuracy between children with right and left UCP. Our results did not show significant differences between children with right and left UCP, thus contradicting the initial hypothesis. Although motor functions are predominantly controlled by the left hemisphere, perinatal brain injury can disrupt the typical course of brain development (Kuo et al., 2017). Children with UCP will not always have a typical contralateral reorganization pattern (Butson et al., 2014). Cortical projection patterns may also rearrange and occur in an ipsilateral or mixed pattern (Staudt, 2010). The lesion in one of the cerebral hemispheres can weaken the contralateral projection while strengthening the ipsilateral cortical spinal tract (Eyre et al., 2007). In this way, the uninjured motor cortex controls bilateral movements, while the injured motor cortex does not control any movement in children with UCP (Kuo et al., 2017). However, this is speculative, as there were no neuroimaging data available in the present study.

It was hypothesized that the functional performance and working memory scores of children with UCP would correlate with higher rates of accuracy and lower RTs in the HLJ task. Results suggest that the functional performance and working memory of children with UCP are associated with their MI performance. Using the AHA task, which is a standardized and valid instrument to measure functional performance (Krumlinde-Sundholm et al., 2007), it was revealed that the upper limb function is a performance correlate in the HLJ task. Although Williams et al. (2011) also reported associations between accuracy in the HLJ and functional performance, the authors were cautious in drawing conclusions since they used a subjective questionnaire applied to the parents to evaluate functional performance. Other findings reinforce the hypothesis that MI is closely related to the motor representations involved in the planning and executing movements. The MI ability in children with developmental coordination disorder varies according to their motor impairment level and healthy children with higher functional performance display better MI task performance (Barhoun, et al., 2019; Williams, Thomas, Maruff, & Wilson, 2008). In addition, there is a direct association between MI ability and planning in healthy individuals (Fuelscher et al. 2016). Children with poor functional performance have limitations in physical movements, resulting in an inability to properly develop internal movement representations (Williams et al., 2012). Taken together, it is possible to suggest that the functional level of a child with UCP affects his/hers MI ability.

The hypothesis that MI would correlate with working memory was corroborated. Similar results were reported in adult patients after stroke (Malouin et al., 2004). Working memory is also involved in learning new motor skills, especially in the early stages of the learning process (Pascual-Leone et al., 1995). The present findings are in accordance with functional neuroimaging studies showing activation of working memory networks during MI tasks (Ruby & Decety, 2001; Deiber et al., 1998). These results suggest that success in MI tasks may depend on the children's ability to maintain and manipulate information in working memory. In fact, to perform MI, patients are instructed to retrieve the kinesthetic sensations of movement contained in working memory in order to choose the most appropriate motor strategy. To the best of our knowledge, this is the first study to examine the association between MI and working memory capacity in children with UCP. Results emphasize the role of cognitive factors on MI and suggest that cognitive deficits should be taken into account when assessing the ability of these children to perform MI tasks.

Some limitations of the study concern certain variables that may affect the ability to perform MI and that were not controlled. It was not possible to evaluate the presence of body perceptual and representational deficits. This type of deficit has been shown to occur in some children with UC P (Fontes et al., 2017) and may negatively influence MI ability (Courbois, Coello, & Bouchart, 2004). In addition, attention and IQ can influence the performance of groups. In our study, it was not possible to assess attention; moreover, due to logistic reasons, IQ measurement was performed only in children with UCP. It is also worth highlighting another methodological limitation of the present study. According to the literature, children can use visual strategies to solve the HLJ task (Steenbergen, van Nimwegen, & Crajé, 2007). One way to prevent them from using visual strategies is to cover the children's hands with a cloth. However, in our study, children's hands were not covered. Despite this, it is improbable that the visual strategy was used since the effects of biomechanical constraints were observed.

The study has relevant implications for clinical practice. Although the findings indicate that children with UCP can perform MI, it must be considered that these individuals constitute a heterogeneous group, with wide-ranging neuromotor disabilities, depending on the location and extent of the neurological injury. Thus, considering that MI training is used in neurological rehabilitation, the child must undergo an individualized assessment of MI ability to ensure the technique's effectiveness. A rehabilitation program based on MI may improve the motor planning of children with UCP, since MI leads to the activation of sensorimotor representations involved in motor planning and execution (Steenbergen, Crajé, Nilsen, & Gordon, 2009). The use of this rehabilitation tool has been poorly explored in children with UCP (Steenbergen, et al., 2009). Using a MI protocol associated with physical practice, Souto et al. (2020) found improvements in bimanual performance to perform daily tasks in children with UCP. The children were asked to watch videos with motor content and then implicitly imagine the assisted motor task, and finally, they physically performed the task (Souto et al., 2020). MI has also been used in rehabilitation through Action Observation intervention. In this approach, participants must watch motor actions and then perform them physically. In the systematic review by Sarasso et al. (2015) most of the reviewed articles showed positive results from action observation in rehabilitation. In a pilot study involving children with UCP, Buccino et al. (2012) showed the effectiveness of treatment by action observation in improving the motor function of the participants. Based on the results of these preliminary studies, it is feasible to suggest that MI is a promising tool in the rehabilitation of the upper limb of children with UCP; however, future studies with larger samples are necessary.

Conclusions

In conclusion, we found that children with UCP injuries on either side of the brain were able to engage in MI to solve the HLJ task. In addition, MI ability is affected by functional performance and working memory. Our results suggest that children with UCP could benefit from rehabilitation programs based on MI. However, as this is an heterogeneous group, with varied neuromotor impairments, it is important that each child with UCP undergoes an individualized assessment of MI prior to the intervention.

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STUDY 4: EFFECT OF MOTOR IMAGERY COMBINED WITH PHYSICAL PRACTICE ON UPPER LIMB REHABILITATION IN CHILDREN WITH HEMIPLEGIC CEREBRAL PALSY.

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Abstract

Introduction: Evidence indicates that motor deficits in hemiplegic cerebral palsy (HCP) impair both motor execution and planning. However, current rehabilitation efforts focus mainly on relieving impairments in motor execution. Motor imagery (MI) is a promising method for stimulating neural networks underlying the planning and control of movements. OBJECTIVE: Evaluate the effectiveness of MI combined with physical practice in improving the function of the upper limbs in children with HCP. Method: Twenty-four participants, aged 7–14 years were divided into two groups: intervention group (IG) and control group (CG). The IG was subjected to MI training and physical practice twice a week for eight consecutive weeks, while the CG received conventional therapy. Participants were assessed with the Assisting Hand Assessment (AHA) at pre-intervention, post-intervention, and follow up. Results: The results showed improved motor functions in both groups. Analysis using the general linear model (analysis of covariance) and Bonferroni post hoc tests showed significant improvements from pre-intervention to post-intervention in the AHA for the IG. The CG showed non-significant improvement in AHA scores. Conclusions: These findings suggest that the MI training, combined with the physical practice program used in this study, was effective in improving upper limb function in children with HCP.

Keywords: motor imagery; rehabilitation; upper limb; children; hemiplegic cerebral palsy

Introduction

In children with hemiplegic cerebral palsy (HCP), the ability to perform various hand activities is reduced. Sensory and motor impairments observed in the affected upper extremities are a major cause of functional compromise (Rosenbaum et al., 2006). These impairments limit performance on simple tasks of daily living, such as changing clothes, brushing teeth, combing hair, feeding, and playing, and can be limiting in a wider social context as well (Rosenbaum et al., 2006). Therefore, an obvious goal of neurorehabilitation is to improve the capacity and performance of the affected arm, in order to promote its effective use in daily tasks (Gordon, 2011). According to Buccino et al. (2012), even when involved in an integral rehabilitation program including conventional physical therapy, use of orthosis, and treatment of spasticity, around 75% of children with HCP may present motor impairments in activities of daily living. Therefore, there is an urgent need to propose new rehabilitation programs that aim at adding to the effects of conventional therapy.

In this study, we assessed the feasibility of using motor imagery (MI) as an adjunct technique to improve upper limb motor function in children with HCP. MI could be an alternative or ancillary approach in these children's rehabilitation of the upper limb. It has been investigated as a therapeutic option in adults with post-stroke hemiplegic deficits (Liu, Chan, Lee, & Hui-Chan, 2004; Crosbie, McDonough, Gilmore, & Wiggam, 2004). To the best of our knowledge, there is no investigation of this approach with HCP.

It has been proposed that motor deficits observed in children with HCP involve impairments in both motor execution and planning (Steenbergen, Verrel, & Gordon, 2007). However, the current rehabilitation techniques focus predominantly on deficits in motor execution and do not specifically address deficits in the movement preparation processes, i.e., in motor planning. Evidence suggests that problems with motor planning can also adversely affect the performance of activities of daily living, and therefore, need to be treated (Steenbergen, Jongbloed-Pereboom, Spruijt, & Gordon, 2013). Research suggests MI as a promising method to train the "cognitive" aspects of motor behavior that can be effective in reducing the motor planning deficits observed in children with HCP (Williams, Reid, Reddihough, & Anderson, 2011; Steenbergen, Crajé, Nilsen, & Gordon 2009).

MI is a cognitive process, in which the covert trial of a motor action is conducted via manipulation of motor representations in working memory without any external manifestation of the motor act (Jackson, Lafleur, Malouin, Richards, & Doyon, 2001). According to Jeannerod (2001), MI is closely related to the motor representations involved in the planning and execution of movements. According to Sirigu and Duhamel (2001), MI corresponds to a process by which the nervous system activates a motor plan and follows its deployment through internal feedback signals, while the motor output remains in a state of inhibition. It is postulated that MI is endowed with similar properties as those of the corresponding motor act and may be involved in the same causal relationship in the generation of a movement (Jeannerod, & Decety, 1995). Thus, when performing MI tasks, impulses are generated and sent to the muscles responsible for that action. This activation may have an important role in assisting the learning and improving of motor skills (Braun, Beurskens, Borm, Schack, & Wade, 2006; Grezes, & Decety, 2001). This hypothesis is supported by a brain imaging study led by Jackson et al. (2001), which demonstrated that MI induced changes in the pattern of brain activation in cortical areas associated with motor planning.

Previous studies have emphasized the similarities between the executed and imagined movements, with regard to neurophysiological and psychophysical parameters, providing evidence that both may be based on similar processes (Malouin, Richards, & Durand, 2012; Jeannerod, 2001; Grezes, & Decety, 2001). From the neurophysiological point of view, experiments using functional magnetic resonance imaging showed that the neural structures activated during the execution of movements are also activated during MI tasks. Specifically, brain regions, such as the supplementary motor area (Grezes, & Decety, 2001), premotor cortex (Jackson et al., 2001), primary motor cortex (Gerardin et al., 2000), cerebellum (Lotze et al., 1999), and posterior parietal cortex (Grezes, & Decety, 2001), are activated during both execution and imagery of different motor actions. Considering the psychophysical similarities, behavioral studies have shown that the time taken to imagine a movement and its effective implementation are temporally coherent (Parsons, 1994). A similarity between MI and execution is also observed with regard to changes in heart and respiratory rate observed during MI tasks. This suggests similar actions of the autonomic nervous system in both situations (Oishi, Kasai, & Maeshima, 2000).

From this evidence, we suggest that MI may favor the acquisition of motor skills through systematic mental trials. Steenbergen et al. (2009) proposed that MI may be useful in training the motor neural networks after injury in the central nervous system. Previous studies mostly investigated the effectiveness of MI in acute (Malouin, Richards, & Durand, 2012) or chronic post-stroke aged patients (Sharma, Pomeroy, & Baron, 2006). Experimental studies indicate a tendency for positive effects of MI on training of motor learning (Jackson Lafleur, Malouin, Richards, & Doyon, 2003), reduction of sensorimotor deficits (Liu et al, 2004), improvement of upper limb function (Page, Levine, & Leonard, 2007), cortical reorganization (Page, Szaflarski, Eliassen, Pan, & Cramer,2009), and performance improvement in the execution of daily activities (Crosbie et al., 2004) in post-stroke subjects. A systematic review by Braun et al. (2006), and a meta-analysis by Kho, Liu, and Chung, (2014) investigated the effects of MI training in the recovery of upper limb function in post-stroke patients. Both studies concurred that MI training effects were beneficial (d=0.5).

Based on these adult post-stroke beneficial effects, it could be hypothesized that children with HCP might also benefit from a rehabilitation program involving the use of MI. In a preliminary small-scale study, Cabral, Narumia, and Teixeira, (2010) evaluated the effects of MI training on three children with diplegic cerebral palsy by assessing their ability to climb a ladder. The results showed major reductions of up to 88.12% in the time needed to perform the task. In a subsequent study, Cabral-Sequeira, Coelho and Teixeira, (2016) evaluated the effects of pure MI training and its combination with physical practice in motor learning of a sighting task that required speed and precision with the paretic arm of children with HCP. In this experimental design, the experimental group (n = 8) underwent 1 day of mental practice and one of physical practice, while the control group (n = 8) underwent recreational activity on the first day and physical practice on the second day. The authors concluded that MI training appears to be a potentially useful feature to increase motor learning in individuals with HCP. The gains obtained can be justified by the fact that the imagined movement modulates the activity in the neural network, increasing the potential of the physical practice to induce higher levels of motor performance (Cabral-Sequeira, Coelho & Teixeira, 2016).

Despite such evidence, the use of MI in the rehabilitation of the upper limbs of children with HCP has not been explored extensively. Consistent with the experimental results of Cabral-Sequeira, Coelho, and Teixeira (2016), MI associated with physical practice seems to be an effective tool in inducing neural plasticity and improving motor performance. It is also believed that for certain motor tasks, imagined movement can lead to higher performance gains than those observed with physical practice (Allami, Paulignan, Brovelli, & Boussaoud, 2008). In addition, another advantage of this method is the non-exclusion of children with limited physical ability, since this is a factor that limits their participation in many rehabilitation protocols. Thus, for children in whom severe motor limitations impede movement, imaginary training can help to keep the motor program active, facilitating the future execution of movements (Lameira et al., 2008).

The present study aimed to investigate the effects of MI associated with physical practice on upper limb improvement in children with HCP. We developed a specific treatment protocol that aimed at training activities of daily living. We used a quasi-experimental, intelligence-controlled study in 24 children with HCP. We hypothesized that children who received MI intervention associated with physical practice would show better results when compared to children in a control group.

Methods

Participants and study design

Children with HCP were recruited from two large university-associated clinical rehabilitation centers located in Belo Horizonte and its metropolitan area in Minas Gerais, Brazil. Twenty-four participants diagnosed with HCP, aged 7–14 years (mean=10.75, SD=2.08) were included in the study. Nineteen children had probable lesions in the left cerebral hemisphere (right hemiplegia) and 5 had probable lesions in the right cerebral hemisphere (left hemiplegia). The individuals eligible to participate in the study met the following inclusion criteria: normal intelligence and working memory, and the ability to collaborate with the physical or occupational therapy at the enrolled institutions and ability to perform the MI task. The ability of motor imagery was assessed by the task of mental rotation (see Steenbergen, van Nimwegen & Craje, 2007). The study excluded children with associated pathologies, such as progressive, epilepsy or hydrocephalus, genetic syndromes, movement disorders, or children who had surgery or botulinum toxin injections in the last 6 months.

The participants were divided into two groups: intervention group (IG; n=12, mean age=10.25, SD=2.95 years) and control group (CG; n=12, mean age=11.25, SD=2.66 years). Group allocation was defined according to geographic location of the participating institutions. Children in the nearest located institution performed the MI training (IG). Children in the other institution served as controls (CG), requiring only the pre-test and post-test assessments, and receiving conventional care in between. IG participants also received conventional treatment regularly.

In a quasi-experimental study, we compared the performance in pre-test and post-test outcome measures of two non-randomly selected groups of children with HCP. One group received MI training, as well as conventional physical therapy (IG); the other group received only conventional physical treatment, and served as control (CG). The primary outcome was measured through the Assisting Hand Assessment (AHA) [41]. The results were controlled for intelligence.

Assessment measures

The domains assessed to select participants and to control for confounding effects were as follows: intelligence, evaluated through the Raven's Coloured Progressive Matrices test (Bandeira, Alves, Giacomel, & Lorenzatto, 2004) and the Block Design subtest of the Wechsler Intelligence Scale (Wechsler, 2002); the working memory evaluated by the backward Digit Span and backward Corsi Cubes tests (Santos, Mello, Bueno, & Dellatolas, 2005); and manual ability according to the Manual Ability Classification System (MACS) (Eliasson et al., 2006).

Main outcome measure

The AHA (version 4.3) was used as an outcome measure to evaluate the effects of MI training on upper limb function (Krumlinde-Sundholm, Holmefur, Kottorp & Eliasson, 2007). The instrument was selected for evaluating the efficiency with which a unilateral disabled child makes use of their (assistive) affected upper limb during activities that require bimanual coordination. First, a 10—15 min play session with a specific toy from the AHA test kit, which requires bimanual manipulation, is video recorded. Later, the video recordings are analyzed based on 22 predefined items by using a classification scale ranging from 1 to 4 points. The sum of the raw score ranges from 22 (low capacity) to 88 (high capacity) points. The instrument has excellent reliability and validity (Holmefur, Krumlinde-Sundholm & Eliasson, 2007; Krumlinde-Sundholm et al., 2007). For statistical data analysis, the raw score obtained by the participants was considered. A licensed physical therapist, familiar with the AHA, conducted the evaluation. Video evaluations were made by a blinded, trained therapist.

Interventions

MI training protocol: This is the first study to assess the effectiveness of MI training in the rehabilitation of the upper limbs of children with HCP. The MI training protocol used in this study was established based on other investigations that used MI on adults with post-stroke hemiplegia (Malouin, Richards, & Durand, 2012; Riccio, Iolascon, Barillari, Gimigliano, & Gimigliano 2010; Page, Levine, & Leonard 2007), and on children with dyspraxia (Wilson, Thomas, & Maruff, 2002).

The activities in which the children had difficulty performing independently were selected to compose the MI training protocol (see Table 1). The performance of the tasks by a 12-year-old girl was recorded and used as a model for both imagined and physical execution. For each task, a video was made lasting between 1080 s.

The training protocol of MI for daily activities was conducted as follows: (1) initially, each participant was instructed to focus on the movement technique of the model performing the task (third-person perspective). (2) Next, the participant was asked to concentrate and try to perform this task mentally on his/her own (first person perspective). (3) After mental training, the participant was required to perform the activity physically; the objects needed to perform the tasks were provided on a table. The combination of MI and physical practice was used in the study in view of the evidence that when MI and physical practice are provided in the same session, the results are synergistic (Malouin, Jackson, & Richards, 2013).

In each session, participants in the IG performed all activities listed in the MI protocol. The sessions took place twice a week, for eight consecutive weeks (Table 1). The average length of the sessions was 50 min and they were conducted in the rehabilitation center that the participant attended. IG participants continued to receive conventional therapy (see details below). The MI training was conducted by the first author, who is a licensed physical therapist.

Table 1. Daily living activities of participants trained in the MI protocol

1	Sharpening a pencil and using it to write
2	Cutting with scissors
3	Holding a cup and bringing it to the mouth
4	Taking a spoon and bringing it to the mouth
5	Brushing teeth
6	Holding and throwing a ball
7	Opening a jar of cookies
8	Putting on shoes and tying laces
9	Putting on a blouse
10	Buttoning a blouse
11	Closing the zipper of pants
12	Combing hair
13	Opening a door knob
14	Using a key to open the lock of a door
15	Turning over the pages of a book

Conventional therapy: The participants of the CG received no MI training. All individuals continued treatment with conventional therapy. The sessions in the rehabilitation centers were offered once or twice a week, according to the children's needs. The care offered to participants in the two rehabilitation centers was similar, since both centers are school clinics in partnership with the same University. The duration of the session averaged 50 min and included muscle stretching, strengthening exercises, and exercises to improve grasp function, manipulation, grip, and fine pinch, among others. In contrast to the MI protocol, which focused on upper limb function, conventional therapy sessions also addressed the recovery of plegic lower limbs through stretching, muscle strengthening, balance and gait training.

Procedures

All research procedures were previously approved by the local research ethics board. Participation was dependent on written informed consent by parents and oral consent by children. Each participant was individually evaluated. The first evaluation (pre-intervention) occurred in the first week of the intervention period. The second evaluation (post-intervention), using only the AHA, was performed at the end of the intervention period and 8 weeks after the intervention (follow-up). Researcher 3, a licensed physiotherapist, performed all assessments blindly. Cognitive tests were performed by a licensed psychologist (researcher 4).

Participants allocated to the IG underwent the MI protocol, twice a week, for eight consecutive weeks with an average of 50 min per session. Throughout the training for the MI tasks, the participants were seated comfortably on a chair that was positioned 50 cm away from a 14 inch flat screen laptop. Each previously recorded video was presented individually on the laptop screen. The participants were instructed to watch the videos attentively, concentrating on the technique used by the model performing the task. After this observation period, the participants were asked to perform the task mentally. The actions were presented in a fixed order according to their complexity, as judged by the experimenters. Each mental trial was repeated five times. At the end of each mental repetition, the objects needed for performing the activities were placed on the table. The participants were then instructed to perform the action as demonstrated in the video.

The participants of the CG continued treatment with conventional therapy, which was offered once or twice a week by a physical or occupational therapist. The treatment goals established by the therapists who tended to the children were maintained and no change of routine care occurred during the study period.

Statistical analyses

Sample homogeneity, in relation to sex, laterality of hemiplegia, and manual ability level, was assessed using the chi-square test. The Student's t-test for independent samples was used to compare the mean age, and the z-score was used for intelligence, working memory, and the AHA prior to the intervention.

Between-group differences in AHA scores in the post-test were analyzed using general linear models (analysis of variance). To investigate to what extent intelligence would influence the results, a model of analysis of covariance (ANCOVA), including intelligence as a covariate, was used. When the results showed significant main effects, the Bonferroni post hoc analysis for multiple comparisons was used to identify differences. The level of significance was defined at p<0.05. Analyses were performed using SPSS (version 1.4).

Results

All 24 participants who underwent the pre-intervention evaluation completed the study. The groups were homogeneous in relation to sex, laterality of hemiplegia, and MACS level of manual ability. The mean age did not differ significantly between the groups. The number of sessions carried out by participants did not differ between groups. Information regarding the participants' characteristics and results of between-group comparisons are provided in Table 2.

Table 2: Characteristics of study participants

Characteristics		Intervention group (n=12)	Control group (n=12)		
		%	%	$\chi 2$	*p*
Sex	Male	7 (58.34%)	6 (50%)	0.168	0.682
	Female	5 (41.66%)	-		
Laterality of hemiplegia	Right	10 (83.33%)	9 (75%)	0.253	0.615
	Left	2 (16.66%)	3 (25%)		
MACS	I	3	4	0.343	0.842
	II	6	6		
	III	3	2		
		Mean (SD)	Mean (SD)	*t*	*p*
Age (years)		10.25±2.95	11.25±2.66	0.871	0.394
Number of sessions (conventional therapy)		12.75±3.22	13.83±2.94	0.859	0.4

MACS: Manual Ability Classification System; SD: standard deviation

The groups did not differ significantly regarding intelligence and working memory, as shown in Table 3. Moreover, both groups started the study with a similar level of upper limb functional performance, as evidenced by the AHA (t = 0.026; p<0.980).

Table 3. Between-group comparison of intelligence and working memory

	Intervention group	Control group			
Intelligence	M (sd)	M (sd)	*t*	*p*	*d*
Raven's CPM or WISC Block Design	-0.648 (0.434)	-0.734 (0.268)	0.582	0.566	-0.24
Working memory	M (sp)	M (sp)			
Digit span	3.83 (0.835)	4.01(0.739)	-0.518	0.611	-0.23
Corsi cubes	3.75 (0.754)	3.92 (0.515)	-0.632	0.534	-0.26

Note: intelligence test values are expressed in z-score. M: Mean; sd: standard deviation

The results of the comparisons between the intervention and control groups based on the AHA scores from pre-intervention to post-intervention and follow-up assessments are provided in Table 4. Although group differences in intelligence were not significant, the effect sizes were non-negligible. Thus, we decided to control for the effects of intelligence in the outcome analysis. The results of analysis of variance (ANCOVA) for the three measurement points indicated a statistically significant interaction between the group and the AHA (F 7.94; p <0,001; η2 = 0.254), when controlled for the effects of intelligence. Post-hoc comparisons indicated that, for IG, significantly higher values were found in the post-test and follow-up compared to the pre-test (p <0.05). Post-intervention and pre-intervention AHA scores differed for the IG (p = 0.001). We also observed significant differences between the pre-intervention and follow-up measures (p = 0.015) in the IG. Post hoc comparisons for the three sequential AHA measures did not differ for CG (p>0.05).

Table 4. Comparison between outcome measures in the intervention and control groups

	Pretest	Post test	Follow-up	ANCOVA			Post Hoc		
	M (sd)	M (sd)	M (sd)	*F*	*P*	*η2*	Pretest x Post test	Pretest x Follow- up	Post test x Follow- up
IG	60.33±15.49	64.42±16.13	63.75±16.04	6.265	0.029*	0.657	0.001	0.015*	0.102
CG	60.17±16.25	61.00±15.98	61.42±15.74	0.151	0.927	0.052	0.101	0.094	0.412

**p*<0.005; IG = Intervention group; CG = Control group; M: Mean; sd: standard deviation.

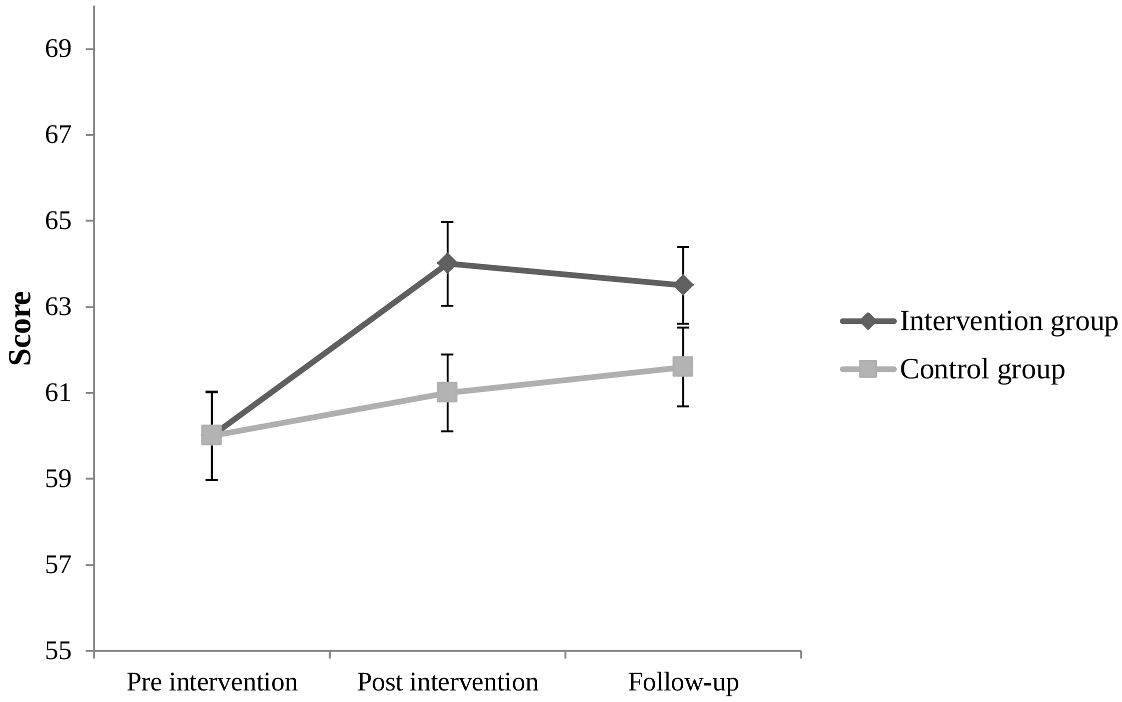

Figure 1: Progression of the evaluation means carried out at three different time points.
Note: Each point represents the average obtained by the groups in the AHA test during pre-intervention, post-intervention and follow-up measurements.

Discussion

In this study, we evaluated for the first time the effect of MI training as a therapeutic alternative, for motor rehabilitation in children with HCP. The results of this study document changes in upper extremity function resulting from a MI protocol associated with physical practice. The IG practiced MI followed by the physical execution of bimanual activities of daily living in two 50-min weekly sessions for 8 weeks. Measures in the CG were taken at corresponding intervals. Significant differences in the AHA were obtained between post-test and pre-test comparisons, and the IG showed a large effect size, even when controlling for the effects of intelligence. Results of the follow-up measure in the IG were non-significant and the effect size was moderate. No intragroup differences for the AHA were observed in the CG, and effect size was small.

Our results suggest that MI training could be a feasible adjunct to physical recovery of upper limb function in HCP children. This is consistent with results of previous studies showing beneficial effects of MI training on upper limb function in adults with stroke sequelae (Kho, Liu, & Chung 2014; Malouin Jackson, & Richards, 2013; Page, Levine, & Leonard, 2007; Liu et al., 2004), walking in diplegic children (Cabral, Narumia, & Teixeira, 2010), manual motor performance in children with intellectual disabilities (Porretta, & Surburg, 1995), and motor learning of the upper limb in hemiplegic children (Cabral-Sequeira, Coelho & Teixeira, 2016).

Our results indicate a trend toward greater gains by combining the imagined movement followed by physical practice compared to conventional single therapy. These findings are consistent with previous results showing that MI training increases gains from subsequent physical practice (Cabral-Sequeira, Coelho & Teixeira, 2016; Allami et al., 2014). We believe that the combined practice reinforced the internal representation of the trained motor act. Allami et al. (2008) showed that physical execution and the sensorial feedback of practice activate different neural networks involved in the executed and imagined movement. Previous studies show that imagined movements share neural substrates similar to the movements executed (Malouin, Richards & Durand, 2012; Grezes, & Decety, 2001; Jeannerod 2001) and induce brain plasticity similar to that obtained with physical practice (Liu et. al., 2004; Page et al., 2009; Jackson et al., 2001). In a neuroimaging study, Zhang et al. (2011) found functional brain changes induced by MI training in the fusiform gyrus, striated body and thalamus. Thus, it is possible that the activation of the motor and somatosensory pathways in both practices favored the acquisition of motor skills, helping to establish and reinforce trained movement patterns. Therefore, from the present and previous results, it is plausible that MI training can explain the superior results of protocols combining MI and physical practice compared with protocols based on physical practice alone (Liu et al. 2004, Page et al., 2009).

In one of the few studies that investigated the effectiveness of imaginary training in hemiplegic adolescents, Cabral-Sequeira, Coelho and Teixeira, (2016) showed, with different kinematic variables, the effect of this technique on performance gains in a goal task. Although a single imaginary training session was used in the cited study, the authors showed that the combination of imaginary and physical practice provided superior performance gains when compared to isolated physical practice. We believe that MI training may result in gains in motor function alone, but it also seems to increase the effects of concomitant physical practice. For Gomes et al. (2012), the practice of isolated imagery is inferior to physical and combined practices, but it is more effective when compared to the absence of practice. According to Allami et al. (2008), when combined proportionally, imaginary and physical practice play an important role in motor gains. On the other hand, there is also evidence that the practice combined with lower rate of imagery practice appears to be less effective in inducing improvements in motor performance (Gomes et al., 2012; Allami et al., 2008).

Our results corroborate the findings already found with MI training in typically developing children. Doussoulin and Rehbein (2011) showed that the benefits of MI training were comparable to those found in physical practice. In a group of 9 to 10 year-olds, the authors reported improvement in movement quality and ball-throwing proficiency. In a sample of healthy adolescents, Hemayattalab and Movahedi (2010) found that MI training followed by physical practice produced significant gains in basketball free throw proficiency. In both studies, the experimental protocol involved the imagined movement and the execution of the same actions in a short time, as proposed in our study. The results of these studies support our findings by showing that imaginary training followed by physical practice is more effective when compared to isolated practice.

We found in the literature a large number of studies evaluating the effectiveness of MI training in upper extremity function after brain injury. For adults with hemiplegia, studies show increased hand and finger movement and relearning of functional tasks after the use of protocols involving MI training (Braun, et al., 2006; Crosbie et al., 2004; Jackson et al., 2003). In a study of people with chronic hemiplegia, Page, Levine, and Stephen (2007) showed that the training of MI, associated with physical practice, resulted in a significant improvement in the movement of the affected upper limb compared to the group that only performed physical practice. The meta-analysis by Kho, Liu, and Chung, (2014), showed that 4 out of the 5 studies analyzed reported significant effects of MI in post-stroke patients. Our study is one of the first to evaluate the effectiveness of MI training associated with physical practice in the rehabilitation of the upper limb of hemiplegic children. The results found are similar to those reported in adult hemiplegia and reaffirm the potential of the combination of physical and imaginary practices in neurological rehabilitation.

Our study revealed significant differences between the pre- and post- intervention measures for the IG, evidencing improvements in functional abilities. The absence of statistical differences between the post-intervention and follow-up measures indicates that the improvements obtained persisted after a period of 8 weeks and did not suffer a decrease after the suspension of the intervention protocol. We believe that the maintenance of the motor gains in the children of the

IG was because of the potential of MI training in triggering specific sensorimotor representations that increase the learning potential of physical tasks in the subsequent period, thus achieving neuroplasticity. Strengthening our hypothesis, evidence suggests that motor imaging increases the excitability of different brain areas associated with motion planning and control (Allami et al. 2008; Sharma, Pomeroy & Baron, 2006; Jackson et al., 2003). Previous studies have also demonstrated the persistence of imaging training effects (Cabral-Sequeira, Coelho & Teixeira, 2016; Debarnot et al., 2009), supporting the hypothesis that when associated with physical training, this technique can induce stable performance gains. From these results, our study supports the hypothesis proposed by Steenbergen et al. (2009) that MI is a potential therapeutic tool for the rehabilitation of individuals with cerebral palsy.

Two main limitations of the present study must be discussed. First, the design of this study was not randomized. Group allocation was geographically based. We have reasons to believe, however, that no substantial sociodemographic differences exist in the target population of the two centers. Both centers are affiliated with the same university and follow similar theoretical and methodological guidelines. In addition, the groups were homogenous for all variables evaluated, including gender, age, laterality of hemiplegia, performance of manual ability, and intelligence. Second, the sample size was small. However, it is worth noting that the CG did not present significant differences in the comparisons between pre-intervention and post-intervention and pre- and follow-up- measures, with a small effect size, $d=0.05$ and $d=0.03$, respectively. Thus, the sample size needed to obtain statistical significance at $p<0.05$ would require 1237 participants (Mackey, & Gass, 2015).

When performing a statistical power analysis for the comparisons between groups in the post-intervention AHA score, we verified that a sample of 310 volunteers was necessary to reach statistical significance, since the magnitude of the effect size was small ($d=0.20$). When comparing the AHA score between groups during the follow-up period, the effect size was found to be even smaller ($d=0.15$) and would require a sample of 1237 participants. Knowing that the incidence of cerebral palsy is around 2 to 3 per 1000 live births in developed countries and 7 per 1000 live births in developing countries (Paneth, Hong, & Korzeniewski, 2006), it is unfeasible to achieve such a high sample count. In addition, to apply an intervention study on such a large sample would require a large team of researchers as well as greater financial resources.

Despite these limitations, strengths of the study to be highlighted include the use of the AHA, a well-validated outcome measure, and the relatively good comparability of the two groups before receiving the intervention. Regarding intelligence, for example, a small effect size of d=0.24, would require a sample size between n=138 and n=310 to become statistically significant at α=0.05 and β coefficient=0.80. We also required that participants had a reasonable working memory and processing capacity to complete the training. Furthermore, the training protocol was easily comprehensible for the children, it was easy to apply, relevant for daily activities, and can be used even with children with major degrees of paralysis.

Although this study provides preliminary evidence of the effectiveness of MI training in children with HCP, some points can be improved to get better results. For future studies, we believe that the protocol of MI activities should be individualized, i.e., defined based on the needs of each child. In the current study, although the protocol has been defined based on the functional limitations presented by the children, all participants were trained for the same activities that were frequent difficulties commonly faced by the group. Finally, increasing the sample size and using a randomized allocation design is necessary for improving study.

Clinical Implications

Considering the results of the present study, we believe that MI training can be applied to clinical practice. We believe that MI is potentially less invasive and intense than other additional methods of treatment. Moreover, for many individuals with lesions in the central nervous system, the execution of certain movements is very difficult, sometimes impossible, which complicates their active participation in the rehabilitation process. Thus, an additional advantage of this training is that it will include participants who are usually excluded from physical training programs owing to their limited physical ability. Another advantage of the application of MI is that it can be used safely, it does not require special equipment or facilities, and it is a simple and low-cost resource (Sharma et al., 2006). Finally, MI can be used at home without professional supervision (or with parental supervision).

Conclusion

Further development of techniques for the recovery of functionality in HCP patients is essential to promote functional independence and to improve quality of life. MI has emerged as a potential alternative for functional rehabilitation and has been known to reduce motor deficits in this population. With this study, we observed that MI training combined with physical practice appears to be a useful and effective method that presents significant results in improving functional performance in children and adolescents with HCP. However, this study only provides preliminary evidence because there is a lack of clinical trials about the use of this therapeutic approach in children with HCP, and established rehabilitation protocols using MI are not yet available. Thus, future studies are needed to establish training protocols that allow consistency in the results.

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GENERAL DISCUSSION

This dissertation aimed to investigate the presence of bilateral neuromotor impairments in children with HCP at different levels of motor integration, and to propose a new therapeutic approach for functional rehabilitation. This chapter will summarize our main findings and discuss its contributions and clinical implications.

In study 1, we proposed a neuromotor exam capable of assessing deficiencies at different levels of motor system integration. We proved that the neuromotor exam proposed here showed good internal consistency, with KR-20 values higher than 0.84, except for the tasks used to assess the extrapyramidal level, which resulted in values close to 0.4. Possibly, the low KR-20 value is due to the small number of tasks selected for this level. We suggest that other tasks that evaluate extrapyramidal signals can be added to the exam, such as stereotyped tasks, dystonic movements, choreiform movements, stiffness, among others (Aravamuthan, & Waugh, 2016; Dubois, et al., 1995; Hawker, & Lang, 1990; Albin, Young, & Penney, 1989). Despite the low internal validity of the extrapyramidal score, by correlating it with the manual ability classification system (MACS), we found significant correlations, not only for the extrapyramidal score, but for the great majority of neuromotor exam tasks at different levels of integration. MACS is a valid and reliable system for classifying how children with CP use their hands when handling objects in daily activities (Eliasson et al., 2006). The correlation of MACS with neuromotor exam tasks leads us to suggest that the neuromotor exam proposed here is effective and valid for identifying the neuromotor deficits observed in children with HPC. In our opinion, this is an important contribution to the CP literature, as we lack instruments to evaluate in a comprehensive way the neuromotor deficits, both in research settings and in clinical settings.

The neuromotor exam applied in Study 1 also confirmed that neuromotor impairments in children with HCP are present in the three levels of brain integration (pyramidal, extrapyramidal, cerebellar). These findings are important because the studies that evaluate the neuromotor function in HCP are limited to identifying the classical pyramidal signs. Although evidence from neuroimaging indicates that children with HCP have lesions in the pyramidal system, i.e., in the corticospinal, unilateral pathways (Jones et al., 2007), our results still indicate the presence of extrapyramidal and cerebellar signs bilaterally. The basal ganglia (extrapyramidal) and the cerebellum are subcortical structures classically considered as motor structures that perform projections for different cortical areas (Middleton, & Strick, 2000). Damage to these structures can result in signs such as involuntary movements, synkinesis, tremor, dysmetria, incoordination, among others (Brooks, & Thach, 2011; Bhatia, & Marsden, 1994), consistent with the results found in our study. According to Middleton, & Strick, (2000), the presence of these symptoms may be related to the interruption of basal ganglia or to the projection of cerebellar areas of the cerebral cortex that are important for the movement control. Therefore, it is possible that focal brain damage in the cortical region may influence the development of other systems, causing additional effects that are more widespread throughout life (Reid et al., 2015; Friel, et al., 2012; Eyre, 2007). These results raise important questions. Instruments capable of assessing neurological symptoms at all levels of motor integration, including extrapyramidal or cerebellar signs, are extremely relevant for accurate clinical evaluation and appropriate selection of the rehabilitation program. We propose a simple, easy-to-apply, low-cost neuromotor exam, capable of indicating neuromotor signals related to different levels of motor integration and assisting in the selection of the most effective interventions.

A second important finding in Study 1 and little explored in the literature, was the discovery of neuromotor impairments in the non-paretic limb in children with HCP. Our results are consistent with the results of previous studies involving children with HCP who reported deficiencies in coordination, fine manual dexterity, strength, and speed of motion in the non-paretic hand (Hawe et al., 2020; Rich et al., 2017; Tomhave, et al., 2015; Arnould, Penta, & Thonnard, 2008). Although ipsilateral motor deficits are not as severe as those of the contralesional arm, they can substantially affect the functional performance of daily life activities. We consider that the presence of bilateral neuromotor deficits in HCP can be explained, partly, by a compromise in motor planning. Deficits in motor planning have been frequently reported in children and adults with HCP (Krajenbrink et al., 2019; Lust et al., 2018; Steenbergen et al., 2013; Chen & Yang, 2007). Rosenbaum et al. (2004) describe motor planning as a computational process that selects a pattern of behavior among many alternatives that allow the successful execution of action. Thus, motor planning is a pre-condition for optimal performance in daily life activities since it allows the individual to act in a predictive and anticipated way (Rosenbaum et al., 2004). In accordance with our hypothesis, the evidence shows that motor planning deficits can impair the bimanual function in HCP (Steenbergen et al., 2013). These findings contribute to a better understanding of motor deficits in HCP and have important clinical implications for the development and proper application of rehabilitation therapies. Therapeutic approaches for children with HCP should include the non-paretic limb and should also include techniques to recover motor planning deficits. Strategies involving biannual use, such as HABIT, have shown positive results for this population (Friel, et al., 2016). To address motor planning deficits, research suggested motor imagery training (Souto et al., 2020) and Action Observation intervention (Sarasso et al., 2015).

Another issue that was investigated in Study 1 and that deserves further discussion, regards the influence of the laterality of the lesion on the results of the neuromotor exams in children with HCP. With the exception of the 9HPT task for the non-plegic hand (dominant hand), the results of the neuromotor exam showed no performance differences between children with left and right hemisphere lesions. This result is intriguing, considering the behavioral and neurophysiological evidence that showed mastery of motor abilities and bimanual coordination for the left hemisphere (Serrien, Ivry, & Swinnen, 2006). Older studies showed that damage to the left hemisphere affected the movement of both arms, while damage to the right hemisphere only affected the contralesional arm (Harrington & Haaland, 1991). However, subsequent studies have reported similar degrees of ipsilateral motor deficiency after left and right hemisphere injuries (Wetter et al., 2005). We believe that, despite the predominance of the left hemisphere for the control of motor functions, early lesions may impair the typical course of brain development. Cortical projection patterns after early lesions may reorganize and develop into an ipsilateral, contralateral or even mixed pattern (Staudt, 2010). Studies involving neuroimaging are still needed for further clarification.

Although MI has been a widely studied cognitive phenomenon in adults, it has been relatively little investigated in early childhood. One of the main objectives of Study 2 was to verify whether 6 to 7 year-old children are capable of engaging in a task that requires the use of the MI ability. This study was of great relevance, given the importance of mental simulation of action for the development of motor control. Furthermore, MI was identified as a promising therapeutic approach in child rehabilitation (Steenbergen et al., 2009). Using the HLJ task, we found that children from 6 to 7 years of age can perform MI. However, our findings differ from those reported by Spruijt, van der Kamp, & Steenbergen (2015), who found that 6-year-old children cannot perform MI. We believe that the difference in results is due to the task employed. Spruijt et al., (2015) used a mental chronometry task. For Spruijt et al. (2015) mental chronometry paradigm seems to be a conservative measure that may underestimate the ability of individuals to use MI. In our study, we used the HLJ task, widely used in the evaluation of MI (Parsons, 1994). Our results are consistent with studies that suggest that mental simulation abilities undergo strong development over 5 to 6 years of age (Funk, Brugger, Wilkening, 2005). From that age on, the ability to integrate visual and proprioceptive references necessary for the execution of the movement is already present (Guilbert, 2018). Thus, it becomes reasonable to think that interventions based on MI can be used in this age group.

With the results of Study 2, we were able to see that the MI ability improves with increasing age. In addition, we found that at 10 years of age, this capacity is equivalent to that of healthy adults. We believe that the age differences found in this study can be interpreted in reference to the various transitions observed in the behavior of children in this age group, revealing a progressive maturation of the central cognitive processes involved in MI that are necessary to program and perform motor actions. The improvements observed in MI according to age are in accordance with what we know about the maturational development of neural networks known to support motor planning (Thibaut, & Toussaint, 2010; Frith, Blakemore & Wolpert, 2000). Studies on the development of motor control indicate that motor planning develops rapidly between 6 and 10 years of age (Thibaut, & Toussaint, 2010; Westenberg, Smits-Engelsman & Duysens, 2004). In general, both MI and motor planning are supported by an interactive neural network that includes the posterior parietal cortex, the premotor cortex, and the cerebellum (Jeannerod, & Johnson-Frey, 2003). The study by Thibaut, & Toussaint (2010) evaluated the development of motor planning in children aged 6 to 10 years and compared their performance with that of healthy adults. In agreement with our results, these authors reported that only 10-year-old children achieved the same level of performance as adults. 10-year-old children could use and integrate all the tips used by adults to perform the proposed motor planning task. Thus, our results reinforce the idea that MI and motor planning are closely related processes and support the hypothesis that motor planning deficits, which occur in individuals with HCP, are related to the difficulty of performing MI.

Based on converging evidence, which shows that motor planning is compromised in HCP, and given its relationship with MI processes (Steenbergen et al., 2009), the main objective of our Study 3 was to assess the ability to use MI in this group. Following previous studies, we used the HLJ task paradigm to assess MI ability (Jongsma et al., 2016; Steenbergen et al., 2013; Williams et al., 2011). Our results point to important findings. First, we are confident that MI was used by children with HCP, as the results of the analysis indicated that hemiplegics used strategies similar to those used by healthy controls. Nevertheless, it is interesting to note that children with HCP made more mistakes than controls. When evaluated together, these results indicate that children with HCP are capable of performing tasks that trigger the use of MI, however, the accuracy with which they do so may vary and may be related to individual factors. These results are in line with a previous study (Williams et al., 2011). We know that children with HCP are a very heterogeneous group, individual differences such as location, size and time of installation and injuries may be influential factors in motor processes, including MI. Thus, it may be necessary for each child to undergo an individualized assessment of their MI ability. Nevertheless, we consider the children in this study as a group, and as such, we have contacted them in order to be involved in MI tasks. These results are a positive finding for researchers interested in examining MI-based intervention programs as a method to improve motor planning and, consequently, subsequent motor function.

In Study 3, we highlighted the hypothesis that functional performance and work memory score would correlate with MI task performance in children with HCP. Our findings confirmed this hypothesis. Regarding the association of MI with functional performance, this correlation was already expected in view of the neuroimaging studies that showed similar neural substrates involved in the execution and MI (Kuo, et al., 2017; Grezes, & Decety, 2001). Our results in children with HCP are consistent with studies involving healthy children. Associations between MI and functional performance, as well as MI and motor planning, have been reported in healthy children, showing that the better motor capacity, the better performance in MI tasks. (Fuelscher et al., 2016; Hoyek et al., 2009). Besides functional performance, MI also correlated with working memory, corroborating our initial hypothesis. Similar results were reported in adult patients after a stroke (Malouin et al., 2004). Although this hypothesis has not previously been tested in children with HCP, it is an important point since deficits in working memory can limit success in MI tasks. This result is in line with the literature, considering that when performing MI tasks, the individual manipulates and retrieves information from working memory (Malouin et al., 2004). In fact, when performing MI, individuals recover the kinesthetic sensations of motion contained in working memory in order to choose the most appropriate motor strategy. As far as we know, this is the first study to examine the association between MI and working memory capacity in children with HCP. Through this association, it is possible to suggest that the level of functional performance of a child with HCP, as well as his/her work memory capacity, affects his/her MI capacity. Thus, our results suggest that both motor performance and work memory should be taken into consideration when assessing the ability of these children to perform MI tasks.

Finally, after clarifying important gaps involving MI in healthy children with HCP, Study 4 allowed us to evaluate, for the first time, the effects of MI training on improving the functional performance of the upper limb of children with HCP. The results reported here are promising, as they show significant gains in the functional performance of the group that received the intervention. The training protocol developed in this study focused on the training of functional tasks of daily life, which the children had difficulty to perform independently. Besides the imagined movement, the child should also physically perform the trained task. We believe that the combined practice, i.e., MI plus physical practice, reinforced the internal representation of the trained motor actions, leading to a better performance. These findings are consistent with previous results, showing that MI training increases gains with subsequent physical practice (Cabral-Sequeira, Coelho & Teixeira, 2016). The gains from MI training can be explained by the fact that MI increases the excitability of different areas of the brain associated with movement planning and control (Allami et al., 2008; Sharma, Pomeroy & Baron, 2006). Although gains from MI training are well established in adult hemiplegics (Kho, Liu & Chung, 2014), our study is one of the first to evaluate the effectiveness of MI training associated with physical practice in the rehabilitation of upper limbs of hemiplegic children. The results found are similar to those reported with adults, and reaffirm the potential for combining physical and MI practices in neurological rehabilitation. Despite this, this study provides only preliminary evidence, given the lack of clinical trials on the use of this therapeutic approach in children with HCP. As the established rehabilitation protocols using MI are not available, future studies are needed to set up training protocols that allow consistency in results.

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GENERAL CONCLUSION

Our interest in this study was to investigate the presence of bilateral neuromotor deficiencies in children with HCP, including MI deficits, in order to propose a new therapeutic approach for functional rehabilitation.

In Study 1, we investigated whether neuromotor deficiencies in children with HCP are observed at different levels of motor integration and in both upper limbs. We showed that children with HCP presented signs of deficiency in the pyramidal, extrapyramidal and cerebellar systems. In addition, we found that, regardless of the laterality of the lesion, children with HCP also present deficiencies in the non-paretic hand.

In Study 2, we evaluated whether children with typical development, between 6 and 7 years of age, could use the MI ability and whether there would be improvements in this ability as age increases. We found that the MI ability is already present at 6 and 7 years of age and that there has been a progressive improvement until age 10. From the age of 10, children's MI ability is equivalent to that of healthy adults.

In Study 3, we were interested in evaluating the ability of MI in children with HCP, also checking whether the laterality of the lesion, functional performance, and work memory could influence their performance. We found that although they made more mistakes than healthy controls, children with HCP may be involved in a task that requires the use of MI. In addition, functional performance and work memory influence the performance of MI in children with HCP. The laterality of the lesion was not a factor influencing MI ability.

In Study 4, we investigated the potential of an intervention protocol involving MI training associated with physical practice in the functional recovery of upper limbs of children with HCP. We found that the group that received the intervention with both MI and physical practice showed significant gains compared to the control group. By this study, we suggest that MI can be an effective therapeutic alternative for motor function recovery in children with HCP.