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

Thalita Karla Flores Cruz

MULTIDIMENSIONALITY AND SPECIFICITY OF NEUROPSYCHOLOGICAL BODY REPRESENTATIONS IN CHILDREN WITH HEMIPLEGIC CEREBRAL PALSY

Multidimensionalidade e especificidade das representações neuropsicológicas do corpo em crianças com Paralisia Cerebral Hemiplégica

BELO HORIZONTE – MINAS GERAIS
SEPTEMBER/2020

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INSTITUTO DE CIÊNCIAS BIOLÓGICAS
PROGRAMA DE PÓS GRADUAÇÃO EM NEUROCIÊNCIAS

FOLHA DE APROVAÇÃO

Multidimensionalidade e especificidade das representações neuropsicológicas do corpo em crianças com Paralisia Cerebral Hemiplégica / MULTIDIMENSIONALITY AND SPECIFICITY OF NEUROPSYCHOLOGICAL BODY REPRESENTATIONS IN CHILDREN WITH HEMIPLEGIC CEREBRAL PALSY

THALITA KARLA FLORES CRUZ

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“Por vezes sentimos que aquilo que fazemos não é senão uma gota de água no mar. Mas o mar seria menor se lhe faltasse uma gota”.

Madre Teresa de Calcutá

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ABSTRACT

One relevant disorder presented by children with hemiplegic cerebral palsy (HCP) is the impairment of body representations. The processing of body-related knowledge comprises three distinct levels of body representations: body schema (BS), body structural description (BSD) and body image (BI). BS provides on-line information about body parts, BSD allows the individual to specify the position and limits of each part of the body and BI contains semantic and lexical information about body parts. Despite the relevance of body representations to action execution, self-representation and social interactions, few studies have systematically explored the impairments of body representations in children with HCP. The main goals of this dissertation are to investigate possible interactions among body representations during the development of BI, and to investigate distinct subtypes of impairments in body representation (especially, selective deficits in body representation levels) in children with HCP. First, a review was conducted presenting the investigation that was carried out on the evidence for the multiple and distinct body representations. To investigate our main goal, three empirical studies were conducted. In the first study, the developmental structure of lexical-semantic body knowledge (operationalized from the fluency tasks of words related to body) in children with typical development was explored. Qualitative analysis of the semantic network of the body parts category suggests an influence of infant sensorimotor development on the development of BI. In the second study, the performance of children with typical development was compared with that of children with HCP to investigate the development of BI in children with HCP. Children with HCP presented a representational profile of BI (as reflected by their performance in the semantic fluency task), which seemed equivalent to that of younger children with typical development. However, they performed significantly worse than children with typical development of the same age. Such findings allow us to suggest that sensorimotor and visual impairments, frequently present in HCP, influences BI development. In the third study, both bottom-up and top-down approaches were performed to investigate whether children with HCP present impairments of body representation in specific levels, and to identify possible selective impairments in body representation. In the present study, we found four main groups of distinct body representation profiles and 22 cases of selective impairment in body representation levels. The findings of this dissertation provide evidence that models of body representation derived from studies of adults are also helpful in enabling the understanding of body representation disorders in childhood. In addition, these results provide an important contribution regarding body representation impairments in children with HCP.

Keywords: Body representation; Body schema; Body structural description; Body image; Hemiplegic cerebral palsy.

RESUMO

O comprometimento das representações corporais é um distúrbio relevante apresentado por crianças com paralisia cerebral hemiplégica (PCH). O processamento do conhecimento relacionado ao corpo compreende três níveis distintos de representações corporais: o esquema corporal (EC), a descrição estrutural do corpo (DEC) e imagem corporal (IC). O EC fornece informações *on-line* sobre as partes do corpo, a DEC permite que o indivíduo especifique a posição e os limites de cada parte do corpo e a IC contém informações semânticas e lexicais sobre partes do corpo. Apesar da relevância das representações corporais para a execução do ato motor, para a auto representação e para as interações sociais, poucos estudos exploraram sistematicamente os comprometimentos das representações corporais em crianças com PCH. O principal objetivo desta dissertação é investigar possíveis interações entre representações corporais durante o desenvolvimento da IC e investigar subtipos de diferentes comprometimentos na representação corporal (especialmente déficits seletivos nos níveis de representação corporal) em crianças com PCH. Primeiramente, foi realizada uma revisão apresentando a investigação realizada sobre as evidências para as múltiplas e distintas representações corporais. Para investigar nosso objetivo principal, três estudos empíricos foram conduzidos. No primeiro estudo, foi explorada a estrutura de desenvolvimento do conhecimento léxico-semântico do corpo (operacionalizado a partir das tarefas de fluência de palavras relacionadas ao corpo) em crianças com desenvolvimento típico. A análise qualitativa da rede semântica da categoria de partes do corpo sugere uma influência do desenvolvimento sensório-motor infantil no desenvolvimento da IC. No segundo estudo, o desempenho de crianças com desenvolvimento típico foi comparado ao de crianças com PCH para investigar o desenvolvimento de IC em crianças com PCH. As crianças com PCH apresentaram um perfil representacional da IC (avaliado pelo desempenho na tarefa de fluência semântica), que parece equivalente ao de crianças mais jovens com desenvolvimento típico. No entanto, elas apresentaram desempenho significativamente inferior ao de crianças com desenvolvimento típico com a mesma idade. Tais achados permitem sugerir que as deficiências sensório-motoras e visuais, frequentemente presentes na PCH, influenciam o desenvolvimento da IC. No terceiro estudo, foram realizadas abordagens *bottom-up* e *top-down* para investigar se crianças com PCH apresentam comprometimentos da representação corporal em níveis específicos e para identificar possíveis comprometimentos seletivos na representação corporal. Neste estudo, encontramos quatro grupos principais com perfis distintos de representação corporal e 22 casos de comprometimento seletivo nos níveis de representação corporal. Os resultados desta dissertação fornecem evidências de que modelos de representação corporal derivados de estudos com adultos também são úteis para permitir a compreensão dos distúrbios das representações corporais na infância. Além disso, esses resultados fornecem uma importante contribuição em relação às deficiências das representações corporais em crianças com HCP.

Palavras chave: Representação corporal; Esquema corporal; Descrição estrutural do corpo; Imagem corporal; Paralisia cerebral hemiplégica.

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LIST OF ABBREVIATIONS AND ACRONYMS

ASP - average shortest path

BI – body image

BS – body schema

BSD – body structural description

CP – cerebral palsy

CRW – correct and repeated words

D – density

DI – diameter

E – edges

EBA - extrastriate body area

FBA - fusiform body area

FFA – fusiform face area

fMRI - Functional magnetic resonance imaging

HCP - hemiplegic cerebral palsy

LHCP – left hemiplegic cerebral palsy

RHCP – right hemiplegic cerebral palsy

N - nodes

OFA - occipital face area

RCPM - Raven's Progressive Coloured Matrices

SFT - sensory/functional theory

SGA – speech graphs attributes

TD – typical development / typically developing

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1. INTRODUCTION

Cerebral palsy is defined as “a group of disorders of the development of movement and posture, causing activity limitation that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain” (Bax et al., 2005). Hemiplegic cerebral palsy (HCP), a cerebral palsy subtype, is characterized by impaired gross and fine motor coordination associated with pyramidal (spastic) disorders that mainly affects the contralesional side of body. Although it is a motor disorder, depending on the affected area of the brain HCP also can be associated with other disabilities, such as cognitive and emotional processes impairments.

One relevant disorder presented by children with HCP is the impairment of body representations. Previous studies (Fontes et al., 2014; Fontes et al., 2017; Frassinetti et al., 2012; Nuara et al., 2019), in one of which the author of the present dissertation participated (Fontes et al., 2017 – see Appendix 1), have investigated impairments of levels of body representation in children with HCP. The levels of body representation investigated were based on a cognitive-neuropsychological model derived from adult neuropsychological studies (Coslett, 1998; Schwoebel & Coslett, 2005; Sirigu et al., 1991).

According to this model, the processing of body-related knowledge comprises several representations, with three distinct levels of representations: body schema (BS), body structural description (BSD) and body image (BI). BS supplies information about the online representations of the body parts, BSD allows the individual to specify the position and limits of each part of the body and BI contains semantic and lexical information about body parts (Coslett, 1998; Berlucchi & Agliotti, 2010; Sirigu, et al., 1991).

The impairments of the different forms of body representation in children with HCP have important functional and clinical implications. Impairments of body representation may explain the phenomenon of "developmental disregard", conceptualized as an inadequacy to use the potential motor functions of the affected upper limb for functional practice in daily life (Hoare et al., 2007; Houwink et al., 2011).

Impairments in BS have received more attention in the literature and were the subject of other investigations in which the present author participated (Souto et al., 2020[a]; Souto et al., 2020[b];

Souto et al., *no prelo*). In one study, BS impairment was investigated in children with HCP through motor imagery, using the hand laterality judgement task (Souto et al., *no prelo*). Although motor imagery has been identified as a promising strategy for the evaluation and rehabilitation of children with HCP, information concerning the development of motor imagery in childhood and adolescence is scarce. Therefore, before investigating whether children with cerebral palsy were able to engage in a motor imagery task, it was first necessary to investigate whether younger children are able to perform hand laterality judgement tasks (Souto et al., 2020[a] – see Appendix 2). In another study, the possibilities of applying motor imagery to the rehabilitation of children with HCP were investigated (Souto et al., 2020[b] – see Appendix 3). The results of these three studies indicates that: i) children from 6–7 years old are able to perform hand laterality judgement tasks (related to BS), and this ability improves as the participants' ages increase (Souto et al., 2020[a]); ii) children with HCP perform hand laterality judgement tasks, but with inferior performance when compared to children with typical development (Souto, et al., *no prelo*); and, iii) motor imagery training is effective in improving upper limb function in children with HCP (Souto et al., 2020[b]).

Some studies have also investigated BSD in children with HCP and observed dissociations between knowledge of self-body parts and the body parts of others (Frassinetti et al., 2012; Nuara et al., 2019). In a task that requires body-part recognition, children with right-brain lesion did not process self-body parts and children with left-brain lesion did not process others' body parts (Frassinetti et al., 2012). Consistent with these results, when asked to make a self-portrait, children with HCP presented upper limb asymmetries; but, when they made portraits of other children, they did not present alterations (Nuara et al., 2019).

The BI of children with HCP has been little reported in current literature. Most studies are older and lack an empirically validated theoretical foundation. For example, Abercrombie & Tyson (1966), based on drawings of the human figure, focused on the emotional aspects of BI. One of the objectives of the present dissertation, therefore, is the investigation of BI in children with HCP from a cognitive-neuropsychological conceptual framework.

A second question investigated is related to the cognitive architecture underlying the different forms of neuropsychological representation of the body. It is debatable, for example, whether models derived from adult neuropsychology can be applied to the body representation of children with HCP. The effects of plasticity (re)organization of the brain, after brain injuries acquired in utero or in the first months of life, are notorious.

Following early unilateral brain lesions, the brain transfers some functions to homotopic areas of the healthy hemisphere. This reorganization is unique to the young brain. In relation to motor functions, ipsilateral motor tracts can be recruited (Krägeloh-Mann et al., 2017; Staudt, 2010[a]; Staudt, 2010[b]). Also, as the representation of the language network is initially bilateral, after early left hemispheric lesions, language functions can be reorganized to the right hemisphere (Krägeloh-Mann et al., 2017; Staudt, 2010[a]; Staudt, 2010[b]). Regarding the somatosensory system, in the case of periventricular brain injuries, children show only a few somatosensory deficits; but, in the case of cortico-subcortical lesions in the middle cerebral artery territory, there is no evidence of primary somatosensory reorganization (Krägeloh-Mann et al., 2017; Staudt, 2010[a]; Staudt, 2010[b]). In this case, children show severe somatosensory deficits.

A criterion for examining the applicability of the cognitive-neuropsychological model to impairments in body representation in children with HCP concerns the observation of impairments in specific, dissociable forms of body representation. Some clinical disorders are associated with a specific patterns of dissociation, which can be interpreted in relation to the common functional architecture (Temple, 1997). A double dissociation occurs when there are two children with developmental difficulties, impaired on two different tasks, A and B: one child is impaired on A but not on B, and the other child is impaired on B but not on A (Temple, 1997). However, the usefulness of dissociations for understanding developmental disorders is questioned in some studies (Bishop, 1997; Karmiloff-Smith, 1998). According to Bishop (1997), during the course of development, the nature of representations may change and dissociations could disappear over time. However, developmental cases should be able to identify meaningful dissociations that reflect the impairments of a specific causal pathway (Castles et al., 2014). The investigation of cases of double dissociation provides an opportunity to explore hypotheses about the nature of an impairment (Castles et al., 2014).

There is some evidence of impairment of specific levels of body representation, constituting double dissociations in children with HCP (Frassinetti et al., 2012; Guedin et al., 2018; Nuara et al., 2019). Right brain damaged children were impaired in processing self-, but not other people's, body parts, whereas left brain damaged children were impaired in processing others', but not their self-body, parts (Frassinetti et al., 2012). Children with HCP drew self-portraits with upper limb asymmetries, but did not present any alteration when they made portraits of other children (Nuara et al., 2019). By evaluating motor dexterity and finger sense in children with HCP and children with diplegic cerebral palsy, it was found that children with hemiplegia presented dexterity impairment in both hands, but finger sense deficit was evident only in the paretic hand when compared with children with typical development (Guedin et al., 2018).

In summary, regarding the dissociations observed in children with HCP, there were dissociations between recognizing self- and others' body parts, and dissociations between finger sense and motor dexterity (Frassinetti et al., 2012; Guedin et al., 2018; Nuara et al., 2019). However, no single, previous study has observed selective impairments related to the three levels of body representation in children with HCP. Thus, another objective of the present dissertation was to investigate the occurrence of specific impairments, restricted to a level of body representation in children with HCP and its possible multiple dissociations.

First, a review was conducted presenting the investigation that was carried out on the evidence for the multiple and distinct body representations. Were described concepts of BS, BSD and BI, the cognitive-neuropsychological model, the neuroanatomical model and neuropsychological disorders related to body representations (especially in children with HCP).

Three empirical studies were conducted, which were presented as articles, two of which have already been submitted for publication. The first study investigated BI, compared to other conceptual domains, in children of different ages with typical development. The second study applied the graph analysis method to investigate BI differences in children with HCP from 7 to 12 years old, compared to children with typical development. Finally, the third study investigated the occurrence of specific impairments of each of the three levels of body representation in children with HCP. All studies are described in the following sections.

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2. AIMS

2.1. Goals

Given the relevance of the assessment of body representation deficits and the development of efficient cognitive and physical therapy training for clinical populations (as children with HCP), our goals are to investigate possible interactions among body representations during the development of BI, and to investigate distinct subtypes of impairments in body representation (especially selective deficits in body representation domains) in children with HCP.

2.2. Objectives

a. To describe the developmental structure of lexical-semantic body knowledge related to BI, in children with typical development (TD);

b. To investigate the development of BI comparing the performance of children with TD with that of children with HCP;

c. To qualitatively investigate whether BS (related sensorimotor information) and BSD (related visuospatial information) contribute to the development of BI;

d. To investigate whether the development of BI is delayed in children with HCP;

e. To investigate if children with HCP present different profiles of impairments of body representation;

f. To identify possible selective impairments in body representation.

3. METHODS

To investigate the main goals, we conducted one review and three empirical studies.

To describe the developmental structure of lexical-semantic body knowledge in children with TD a study was conducted with 204 children with TD aged 4 to 12 years (4-6 years, n=69; 7-9 years, n=59; and 10-12 years, n=76), 56% female, cross-sectionally assessed using word fluency tasks (body parts, foods, animals). BI, operationalized from the fluency tasks of words related to body, was analyzed across age groups using graph network analysis. General cognitive abilities were also assessed.

To investigate the development of BI comparing the performance of children with TD to that of children with HCP, we evaluated 53 children with HCP (age range 7-12 years; mean age = 10.19 [sd=1.83] years; 36 right hemiplegic cerebral palsy and 17 left hemiplegic cerebral palsy) and 204 children with TD (control children, age range 4-12 years, mean age = 8.09 [sd=2.60] years) to qualitatively evaluate whether and how BS (related sensorimotor experiences) and BSD (related visuospatial experiences) affect the development of children's BI, and whether this development is delayed through HCP. General cognitive abilities and the spontaneous production of words (animals and body parts) were assessed by applying the semantic word fluency task. Graph analysis was used to create a lexical-semantic map of body representation from data of a semantic word fluency task.

To investigate if children with HCP present impairments of body representation in specific domains and to identify possible selective impairments in body representation, the performance of 73 children with HCP (age range 5-16 years, mean age=9.03 [sd=2.48] years; 39 right hemiplegic cerebral palsy and 34 left hemiplegic cerebral palsy) in tasks assessing body representation was compared to that of 141 children with TD (age range 5-13 years, mean age=8.17 [sd=1.82] years). General cognitive ability, motor dexterity, and body representational tasks evaluating BS, BSD and BI were applied. Two strategies were employed to identify possible selective impairments in body representation: multivariate classification at the group level (bottom-up approach) and manual single-case identification (top-down approach). Finally, the results of the two analytical approaches were compared.

4. RESULTS

Results will be presented in four sessions: one review and three empirical studies.

Review: Neuropsychological body representations: a narrative review

Study 1: Semantic-lexical knowledge of body parts in typically developing children: graph-analysis of word fluency tasks, under review in *Frontiers in Psychology*, section Perception Science.

Study 2: Body experience influences lexical-semantic knowledge of body parts in children with hemiplegic cerebral palsy, submitted to *Cognitive Neuropsychology*.

Study 3: Selective impairment of body representation domains in children with hemiplegic cerebral palsy: a bottom-up classification approach

On the next sessions, we will present these studies in detail and, afterwards, there will be a discussion chapter summarizing our main findings, describing our limitations and suggestions for futures studies, as well as the main clinical and research implications.

4.1. Neuropsychological body representations: a narrative review

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Abstract

Body representation is a very special form of cognition, considered as the consequence of body experiences. Corporeal awareness refers to perception, knowledge and evaluation of one's own body as well as of other bodies. Based on functional differences, body representations are conceptualized into three levels. This review describes: i) the levels of body representation according to the neuropsychological taxonomies; ii) the cognitive-neuropsychological model of body representations; iii) cortical areas specialized for the processing of body representation; iv) awareness disorders; and v) body representation impairments in children with hemiplegic cerebral palsy. Finally, it is considered several points for future research.

Keywords: Body perception; Body representation; Body schema; Body structural description; Body image.

Introduction

Perception and representation of our own body, based on somatosensory-motor experience, is crucial for action execution, self-representation and social interactions (Berlucchi & Aglioti, 2010). Bodily experience is a phenomenon derived by the combination of different information, such as visual, somatosensory, motor, and proprioceptive inputs and brain regions (Berlucchi & Aglioti, 2010; de Vignemont, 2010; Head & Holmes, 1911; Longo & Haggard, 2012[a]; Longo & Haggard, 2012[b]). In addition, body representation is considered as the consequence of bodily experience and cognition, whereas cognition shapes the body as much as the body shapes the mind (Baumard & Osiurak, 2019).

Body representation levels

According to the triadic taxonomy (de Vignemont, 2010) it has suggested the existence of three body representation domains: body schema (BS), body structural description (BSD), and body image (BI) (Schwoebel & Coslett, 2005; Sirigu et al., 1991). Close to the classical notion of postural schema hypothesized by Head and Holmes (1911), BS is a representation domain derived from sensory input (including muscle, proprioceptive, cutaneous, vestibular, tactile, visual, and auditory) to provide an on-line, real-time representation of one's own body in space (Coslett, 1998). Due to proprioceptive and sensory–motor interactions, BS is essential to the performance of routine motor acts (Coslett 1998; de Vignemont, 2010; Dijkerman & de Haan, 2007; Gallagher, 2005; Paillard, 1999; Rossetti et al., 1995).

BSD is a representational domain composed of the category-specific visuospatial representations of an individual's own body and bodies in general primarily based on vision, but also on somatic perception (de Vignemont, 2010). This representation provide a "structural description of the human body" because it is related to the position of body parts over the body surface, the proximity relationships between body parts and their boundaries (Coslett, 1998; Sirigu et al., 1991).

In some research, BI was referred as all the other representations about the body that are not used for action, whether they are perceptual, conceptual or emotional (body percept, body concept and body affect, Gallagher, 2005). In this thesis, we will use the term "body image" referring to body-related conceptual knowledge (Coslett, 1998). According to this, BI is a domain containing lexical and semantic representations relative to the body, describing the functional purpose of body parts (de Vignemont, 2010).

Cognitive-neuropsychological model of body representations

All those definitions are in agreement with the systematic cognitive-neuropsychological description of body representations based on multiple sensory afferents proposed by Sirigu et al. (1991). This cognitive-neuropsychological model suggests that the processing of body-related knowledge comprises several representations (Figure 01). This model provides a better comprehension about the types of representations and processing necessary to perform tasks involving body representations.

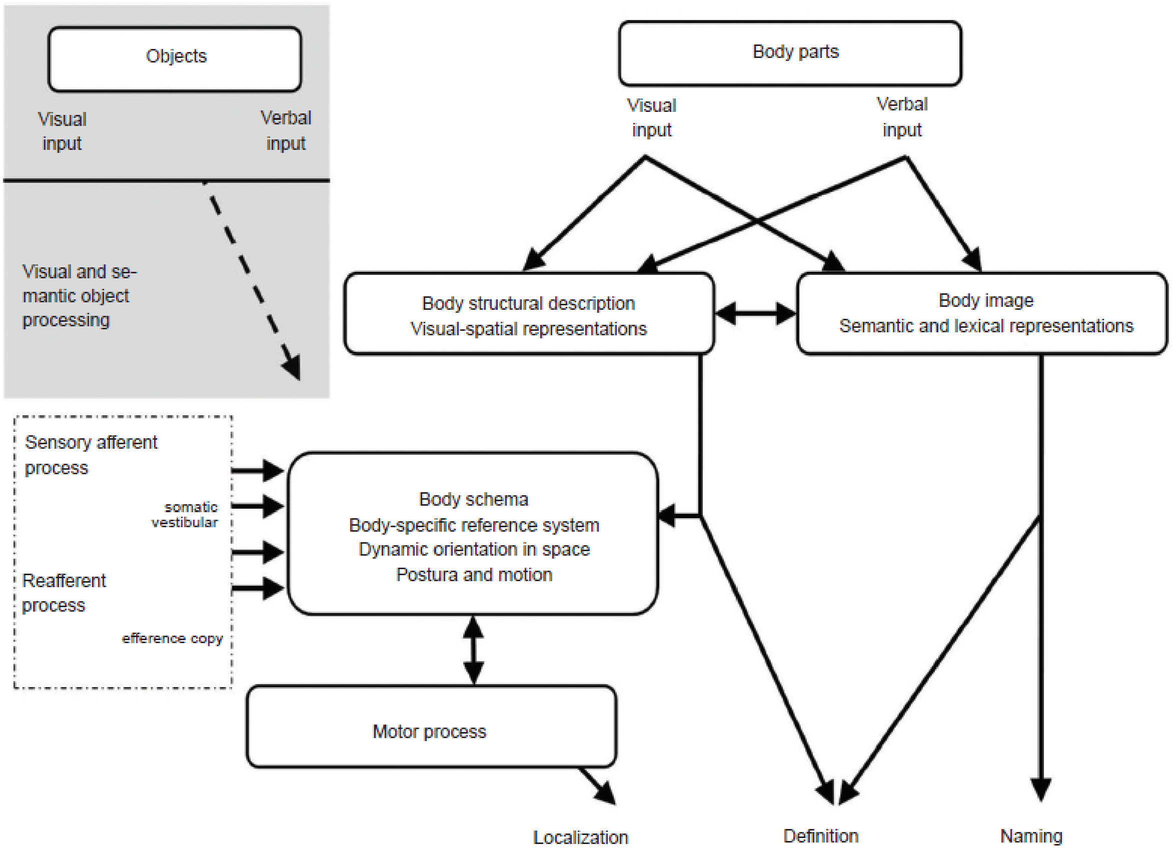

Figure 1. Schematic representation of cognitive-neuropsychological model comprising three levels of body representations (Fontes et al., 2014) adapted from Sirigu et al. (1991).

According to Sirigu et al. (1991), the three levels of body representation system are hypothesized to be independent but can also interact with one another and this interaction of level in tasks involving body parts depend on particular task demands. In a general manner, the information about the actual location of the body segments (i.e., BS) is necessary in hand laterality judgement task. To localize a body part, by a pointing gesture or a verbal response, is mediated by the visuospatial representations (i.e., BSD). Finally, to name and to define the function of a body part requires the semantic and lexical representations (i.e., BI).

Neuroanatomic model of body representations

Studies with adult patients with acquired brain damage and studies of functional neuroimaging have contributed to the growing knowledge of the implementation of the neuroanatomical bases of the three different types of body representation. Regarding their neuroanatomic substrates, processing of body awareness tend to activate mainly three cortical regions: the posterior parietal cortex, the anterior insula and the extrastriate body area (EBA) (Berlucchi & Aglioti, 2010).

Parietal lesions are related to modifications in the representational aspects of gestures and in evaluating and comparing internal and external feedback about movement, suggesting an impairment related to BS (Sirigu et al., 1999). During a task of imitation of meaningless gestures, related to BS, Chaminade et al. (2005) found an activation in the inferior parietal gyrus bilaterally with a specific involvement of the parietal operculum in the left hemisphere. In addition, due to the involvement of visual perception for imitation, increased bilateral occipitotemporal activity was observed (Chaminade et al., 2005). According to Decety et al. (1997) observation of meaningful gestures chiefly activates a left hemisphere frontal network, while meaningless gestures activate the right occipitoparietal areas connected with premotor cortex and also regions within the ventral pathway (cuneus and the inferior temporal gyrus).

In addition, damage to the left temporal lobe was found to be most consistently associated with impaired performance on BSD and BI (Schwoebel & Coslett, 2005). Thus, research about the activation of brain regions for body awareness verified the existence of an association between motor and visual representations (Peelen & Downing, 2007). Self-recognition is also related to the activation of specific areas in the right anterior insula and in the right dorsal cingulate gyrus (Devue et al., 2007). The anterior insular cortex provides a neural substrate that instantiates all subjective feelings from the body and feelings of emotion in the immediate present (Craig, 2009).

Downing et al. (2001) observed that EBA (a region in human lateral occipitotemporal cortex) responds to visual images of human bodies and body parts. In addition to this visual recognition function, the EBA integrates visual, spatial attention, and sensory motor signals involved in the representation of the observer's body (Astafiev et al., 2004). The EBA is involved in the perception of whole bodies and body parts (Dowing et al., 2001; Urgesi et al., 2004).

Functional magnetic resonance imaging (fMRI) studies have also identified a region in the lateral posterior fusiform gyrus sensitive to visual depictions of the human body, the fusiform body area (FBA; Peelen & Downing 2005; Peelen et al. 2006; Schwarzlose et al., 2005). FBA is involved in processing whole body forms in contrast to body parts (Taylor et al., 2007; Taylor & Downing, 2011). The EBA and FBA jointly create a detailed but cognitively unelaborated visual representation of the appearance of the human body (Downing & Peelen, 2011). This representation makes explicit the aspects of the image that contain bodies or body parts, and represents their shape and posture in some detail (Downing & Peelen, 2011). In addition, the fusiform face area (FFA), which is found on the lateral fusiform gyrus, respond selectively to faces (Peelen & Downing 2005; Peelen et al., 2006; Schwarzlose et al. 2005). Another region selective to faces is the occipital face area (OFA), localized the inferior occipital gyrus (Puce et al., 1996). OFA is activated preferentially during the presentation of parts of the face, such as the eyes, nose, and mouth (Pitcher et al., 2007).

Figure 2 presents the brain regions that have attracted more attention as possible specialized sites in different aspects of body awareness.

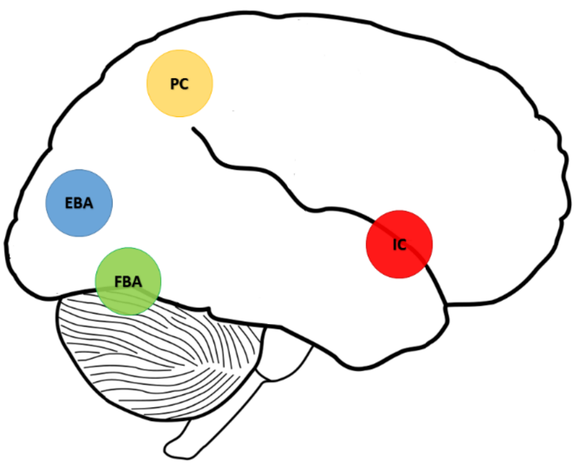

Figure 2. Schematic representation of main cortical regions related with body representation levels (modified from Berlucchi & Agliotti, 2010). *PC = posterior parietal cortex; EBA = extrastriate body area; FBA = fusiform body area; IC = insular cortex.* These cortical regions are present in both cerebral hemispheres, but here they are shown in the right hemisphere because there are evidences for a right-sided dominance for body representations (Berlucchi & Agliotti, 2010).

Awareness disorders

A dysfunction in one or more representational levels (caused by a lesion in some neuroanatomic substrate of body awareness or caused by a psychiatric disorder) could be associated with some bodily awareness disorders. Some examples are allodynia (pain due to a stimulus that does not normally produce pain), anorexia and bulimia nervosa (eating disorders), anosognosia (lack of awareness of one's deficits like hemiplegia), body-specific aphasia (loss of lexical knowledge of body parts), phantom limb (awareness of an amputated limb) or motor neglect (underutilization of one side of the body) (de Vignemont, 2007, de Vignemont, 2010).

Sirigu et al. (1991) reported a case of a patient with autotopagnosia (characterized by inability to localize one's own body parts) who could not localize self or others-body parts either on verbal or nonverbal command, but could name body parts. In agreement with Sirigu et al. (1991), Buxbaum & Coslett (2001), reported another case of a patient with autotopagnosia with severely deficient in pointing to body parts on command or imitation, but with intact BS and BI. These findings suggest that autotopagnosia may be attributable to a selectively on impairment in BSD.

Ideomotor apraxia is a disorder of complex movement characterized by spatiotemporal errors in tool use, gesture pantomime, and/or gesture imitation. Buxbaum et al. (2000) investigated a patient with apraxia who presented deficits in gesture pantomime, recognition, and imitation. The authors concluded that those deficits are related to deficits in dynamic coding of the intrinsic positions of the body parts of self and others, related to BS (Buxbaum et al., 2000). Despite that, Goldenberg (1995) suggests that also the conceptual knowledge about body parts is affected in ideomotor apraxia and ague that basic disorder concerns the relationships between body parts and object function. A disruption of the BS contributes to ideomotor apraxia, but most probably it is only partial, with a concomitant disorder of the BI (de Vignemont, 2010).

Personal neglect is clinically defined by a lack of exploration of half of the body contralateral to the damaged hemisphere. However, neglect patients do not perceive this disturbance. Coslett (1998) found that patients with neglect exhibit an impairment in the BS related to affected side of the body and that the impairment of BS may result in a loss of topographic knowledge of body parts (related to BSD). Also, de Vignemont (2010) ague that there is also a deficit of directing attention to the affected side of the body and this attentional deficit must have consequences on BS and BSD.

Regarding the underutilization of one side of the body, adults with hemiparesis following stroke commonly avoid or suppress the use of the affected limb and learn a strategy compensation with the unaffected limb, a phenomenon named “learned non-use” (Taub, 1980). Similarly, children with hemiplegic cerebral palsy (HCP) develop strategies to be more efficient and effective during their daily lives using non-paretic hand. This “failure to use the potential motor functions and capacities of the affected arm and hand for spontaneous use in daily life” is named developmental disregard (Hoare et al., 2007; Houwink et al., 2011).

Body representation impairments in children with HCP

In general, cerebral palsy (CP) originates from a non-progressive disturbance of the brain that occurred in the developing fetal or infant brain (Bax et al., 2005). This brain trauma around birth generates disorders of the development of movement and posture, often accompanied by disturbances in sensation, perception, cognition, communication and behavior (Bax et al., 2005). Since CP patients could present impaired visual and proprioceptive information, it is suggested that they experience their body and the environment in an unstable perception-movement system (Straub & Obrzut, 2009).

Therefore, children with HCP could present impaired visual and proprioceptive information and frequently exhibits developmental disregard phenomenon. It is also known that body representation domains plays a crucial role in the execution of movements, the recognition of position of body parts, the relationships between body parts and the functional purpose of body parts. Taken together those evidences, it is considered that children with HCP could present impairments in body representations (de Ajuraguerra, 1969; Fontes et al., 2014).

Deficits in body and motor representations in children with HCP have been widely investigated using motor imagery tasks (related to BS – Craje, et al., 2010; Jongsma et al., 2016; Lust et al., 2016; Molina et al., 2015; Mutsaarts et al., 2007; Steenbergen et al., 2007; Steenbergen et al., 2013; Williams et al., 2011; Williams et al., 2012). According to those studies, when compared to typically developed children, children with HCP perform worse in BS tasks (Jongsma et al., 2016; Mutsaarts et al., 2007; Steenbergen et al., 2007). In addition, the ability to execute motor imagery tasks could be related to the involvement of the affected hand, where the less affected hand is able to execute the task, but not the affected hand (Jongsma et al., 2016). In an fMRI study, Chinier et al. (2014) documented specific patterns of brain responses in children with HCP, highlighting that left brain damage affected the execution of motor imagery tasks more than right brain lesions.

Notably, most studies have focused primarily on motor imagery in hemiplegic patients, but body representation impairments in children with HCP extend beyond motor imagery and affect perceptual, semantic, and motor levels of body representation (Fontes et al., 2014; Fontes et al., 2017). Brain damaged children showed a double dissociation in a recognizing body-parts task (related to BSD), whereupon right brain damaged children were impaired in processing self but not other people's body parts, whereas left brain damaged children were impaired in processing others' but not self -body parts (Frassinetti et al., 2012). Also, children with HCP presented upper limb asymmetries when made self-portrait, but did not presented alterations when made portrait from other children (Nuara et al., 2019). Disorders of body schema, body structural description and body image occurs in children with HCP, whereas HCP children perform poorly across several body representational tasks compared to TD children (Fontes et al., 2017).

While there is growing evidence for body representation impairments in children with HCP, an important limitation of the current literature is that few studies have investigated disorders of BI in HCP. In adults without neurological disorders, the processing of words related to body parts results in an increasing activation in the inferior parietal lobe, associated with body perception and postural awareness (Rueschemeyer et al., 2010). Consonant with this, processing of words semantically related to actions (e.g. citing "finger" and "grasping") seems to facilitate movements' execution, by pre-activating a part of the movement circuit (Shebani & Pulvermüller, 2018). Not only related to BS, body semantics are strongly linked to a detailed visuo-spatial body representation, or BSD (Van Elk & Blanke, 2011). The implicit knowledge about the position of body parts is required when processing body semantics (Struiksma et al., 2011). This relation is more pronounced when given a body-related instruction for an action (e.g. requesting that patient comb his own hair), because the hand must find the comb and take it to his the head to comb his hair (Rueschemeyer et al., 2010). Therefore, BI deficits could reflect in success during task practices, frequently applied during physical therapy session (for example).

Children without neurological disorders present better accuracy when naming facial body parts when compared to other parts of the body; also, they present better accuracy when naming body parts related to action when compared to other body parts (Auclair & Jambaque, 2015). This pattern of results allows the hypothesis that BSD and BS shapes BI, suggesting a possible interaction among the different body representations in childhood. Hence, it is important to investigate the hypothesis that BS influences the development of BSD and, consequently, of BI.

This could contribute in understanding the mechanisms underlying the development of BI related to the development of sensorimotor functioning. Further, no one study has used a free naming of body parts task (as in word fluency task) to investigate the development of BI. In addition, little is known about whether and how body representations of children with HCP develop compared to children with TD.

There are only few investigations of how an injury to the immature brain may impact the development of BI during childhood, but no one investigated the body semantic knowledge in HCP. Whether BS and BSD contribute to the development of BI, it is possible to suggest that this development is delayed in children with HCP. More specifically, it allows investigating possible associations of body semantic knowledge and possibly impairments in sensorimotor development.

Nerveless, as mentioned above, evidence indicate that children with HCP present deficits in all the three body representations, regardless of the brain damage laterality (Fontes et al., 2017). However, considering the clinically heterogeneity of disorders presented by children with HCP, it is possible that also exist distinct subtype's of impairments in body representation in children with HCP. Guedin et al., (2018) verified that children with hemiplegia presented dexterity impairment in both hands but finger sense deficit was evident only in their paretic hand. According to the authors, this result change the common assumption that children with HCP who presents satisfactory sensory function also presents good motor outcomes (Guedin et al., 2018). Therefore, it reinforces the hypothesis of distinct subtypes of impairments in body representation in children with HCP. Also, the dissociation among body representations, more precisely in BSD related to others' *versus* self -body parts, were reported in children with HCP (Frassinetti et al., 2012; Nuara et al., 2019). Taken together, these findings could also suggest that children with HCP could present selective deficits in body representation domains, as was observed in adults.

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4.2. Semantic-lexical knowledge of body parts in typically developing children: graph-analysis of word fluency tasks

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Abstract

The literature suggests that body image development is influenced both by body schema and body structural description. We used lexical-semantic knowledge of body parts as a proxy for body image to investigate similarities and differences with other semantic domains (foods, animals) in children of different age groups. We aimed to explore the hypothesis that sensorimotor and visual inputs influence body image development. 204 children aged from 4 to 12 years (4-6 years, n=69; 7-9 years, n=59; and 10-12 years, n=76), 56% female, were cross-sectionally assessed using word fluency tasks (body parts, foods, animals). Lexical-semantic knowledge in these domains was analyzed across age groups using graph network analysis, ANOVA and qualitative analysis of network structure. A similar age pattern for word fluency network metrics occurred across domains (body parts, foods, animals). This included a statistically significant ($p<0.001$) increase of nodes and edges and a decrease of network density for all semantic domains: children aged 10-12 years outperformed children aged 7-9 years, who outperformed children aged 4-6 years. Qualitative analysis of the network structure indicated that with increasing age, words were added to the network nucleus of the previous age group, suggesting cumulative vocabulary development. However, no particular pattern of clustering around distinct semantic features emerged for the foods and animals domains. In contrast, for the body parts domain, children aged 4-6 years mainly identified structures of the head and face, arms, hands, legs and feet; children aged 7-9 years added the joints; and, children aged 10-12 years also identified the internal organs, sub-components of the limbs and axial structures. Lexical-semantic knowledge in different domains presents similarities and differences across age groups. However, the network structure of body-part knowledge presents specific qualitative characteristics suggesting influence of sensorimotor (body schema) and visual (body structural description) inputs on lexical-semantic knowledge (body image) development.

Key-words: Body image; Body representation; Children; Development; Word Fluency; Graph Analysis.

Introduction

Body representation and body knowledge play an important role in several psychiatric and neurologic disorders such as anorexia nervosa (Gaudio & Quattrocchi, 2012; Urgesi et al., 2010; Urgesi et al., 2014), body dysmorphic disorder (Longo, 2015), hemiplegia following early (Houwink et al., 2011), and acquired hemiplegia (asomatognosia, anosognosia - Adair et al., 2003; Baier et al., 2004). The concept of body awareness refers to the perception, knowledge and evaluation of one's own body and the bodies of others (Berlucchi & Aglioti, 2010). The brain employs different sensory channels and different central representations for each of these aspects (Berlucchi & Aglioti, 2010; Goldenberg, 2002).

Several mechanisms coincide within the body, and there isn't one, single map that represents the body (Goldenberg, 2002). Brain lesion and developmental research suggest three main forms of body representation: body schema (BS), body structural description (BSD) and body image (BI) (Buxbaum et al., 2000; Buxbaum & Coslett, 2001; Corradi-Dell'Acqua & Rumiati, 2007; Coslett et al., 2002; Coslett, 2014; Schwoebel & Coslett, 2005). BS is characterized by implicit representations continually updated with movement and adaptation to changes in body properties that synchronize with the motor systems in motor control (Coslett et al., 2002; Haggard & Wolpert, 2005). This provides an "online" representation of the body's properties in space. BSD (visual-structural or topographic description) refers to the topographic representation of the body, providing information about the shape and contours of the surface of the body as well as the relationships among different parts of the body (Coslett et al., 2002). In contrast to BS, which seems to be derived from multiple sensory and motor inputs, BSD is postulated as deriving mainly from visual input (Buxbaum & Coslett, 2001). Finally, BI includes lexical-semantic information about the body such as names for the body parts, associations among body parts and artifacts, and functions of different body parts (Buxbaum & Coslett, 2001; Coslett et al., 2002). Several research lines are concerned with the existence of these multiple and distinct body representations (Buxbaum & Coslett, 2001; Coslett et al., 2002; Head & Holmes, 1911, 1912; Sirigu et al., 1991).

However, although they are distinct, components of the body representation system interact with each other, with some interdependence among them, regarding the development of the different representations (Dijkerman & de Haan, 2007; Sirigu et al., 1991). Studies suggest that the three knowledge levels of the human body in adults (sensorimotor, visuospatial, lexical-semantic) are acquired at different developmental stages (Assaiante et al., 2014; Dijkerman & de Haan, 2007; Slaughter et al., 2004).

Development of body representation knowledge in children

Developmental research has focused mainly on development of BS and BSD in infancy (Bahrick & Watson, 1985; Morita et al., 2012; Rochat & Morgan, 1995; Slaughter et al., 2002; Slaughter et al., 2012). Studies that have assessed development of BS examine body knowledge as providing opportunities for kinetic-visual correspondence. For example, children at four months look and smile more while watching videos of other babies than videos of themselves (Smolak, 2011). Children at five months are able to discriminate between movements of their own legs displayed in a mirror and movements performed by other children (Bahrick & Watson, 1985).

In the study by Rochat & Morgan (1995), children were presented with videos (in real time) of the movements of their own legs from first- and third-person perspectives. Children aged 3 to 5 months tended to look longer at videos from the third-person perspective, probably because there was an image that was visually incongruous with their proprioceptive perception (Rochat & Morgan, 1995). These patterns demonstrate an ability not only to coordinate visual information and motor behavior, but also consistently to integrate sensorimotor information in order to compare images of themselves and others.

Results from Rochat and Morgan (1995) support Rochat's (2010) study. This suggests that children discriminate their own body sensations and experiences from those of others starting at two months. At 12 months, they are able to use the experience of their own bodies to perceive and interpret movement (Morita et al., 2012). However, it is only at 21 months that children begin to recognize or identify themselves as the authors of their own actions (Rochat, 2010). Efficient sensorimotor representations of the body itself (related to BS), which remain throughout life, are expressed at three years (Rochat, 2010).

Regarding the development of BSD, evidence suggests that visuospatial knowledge of the human body begins during the first year of life. Slaughter et al. (2002; 2012) observed that children under 18 months distinguish abstract images of bodies (arms connected to the pelvis and legs that continued to the ears, for example), compared to realistic images of bodies.

Witt et al. (1990) conducted a study on the development of identification of body parts by children from 11-25 months, in which the participants were asked to point out twenty different body parts on a doll. The results showed that, up to 12 months, only a minority of children were able to locate some body parts correctly, and those parts were located on the face. At 15 months, in addition to facial structures, the first parts of the body that children located correctly were the arms, hands, fingers, legs, feet and belly. The ability to locate other parts of the body increased with age. The children were able to locate joints and less prominent structures (for example, neck) only after 24 months. However, this age advantage for locating parts of the body remained for the structures located on the face (Witt et al., 1990).

In summary, efficient BS which remains during life is expressed at three years; but, from five months on, children are able to integrate sensorimotor information in order to compare movements of themselves and others (Bahrick & Watson, 1985; Rochat & Morgan, 1995; Rochat, 2010). Regarding the development of BSD, evidence suggests that visuospatial knowledge of the human body begins during the second year of life and improves with age (Slaughter et al., 2002; Slaughter et al., 2012; Witt et al., 1990).

The number of studies focusing on the lexical-semantic representation of the body, or BI, in older children is increasing. Semantic and lexical knowledge of the body emerges beginning in the second year of life (Slaughter et al., 2004). Camões-Costa et al. (2010) asked children, from 2 years to 3 years and 6 months, to name body parts identified by the examiners. The younger children were not able to name most of the body parts identified. The body parts named correctly correlated with the sensory representation of Penfield's Homunculus (Penfield & Boldrey, 1937). Furthermore, children named parts of the body located on their face with the same accuracy as they named arms, hands, legs and feet. Statistically significant differences were observed between the accuracy of naming the parts located on the face, and the accuracy of naming trunk structures and joints (Camões-Costa et al., 2010).

Auclair & Jambaqué (2014) investigated the influence of visuospatial knowledge (BSD) on lexical-semantic knowledge of body parts (BI) among children aged 5 to 10 years, divided into five age groups. The children had to point to human body parts on pictures made by the examiners. All children showed greater accuracy when naming parts of the body located on the face and parts of the body related to actions, as compared to other parts of the body. It was found that the visuospatial representation of the body influenced the lexical-semantic process, and this influence was not limited to the younger children (Auclair & Jambaqué; 2014).

Crowe & Prescott (2003) conducted a study of 155 children, aged between five and ten years, using the free naming of body parts method. Using cluster analysis, the authors found that the body parts are arranged in topological form (i.e., they are arranged according to their structural proximity). For the older children, this organization is also given a functional form (Crowe & Prescott; 2003).

The findings point to the hypotheses that: 1) the parts of the body that receive more sensory stimuli from the first days of life (such as mouth, eyes, nose and ears) and are used to explore the environment (such as arms, hands, legs and feet) are learned first; 2) with the subsequent acquisition of mobility by children (providing more tactile, kinesthetic, proprioceptive and vestibular experiences), the lexical-semantic learning of the joints is favored; and, 3) visuospatial information also influences the acquisition of lexical-semantic knowledge of the body, thus, learning the dorsal and internal organ structures occurs later during child development.

So, reviewing the literature mentioned above, we formulated the hypothesis that BS influences the development of BSD and, consequently, of BI. The two main goals of the present study are: 1) to describe the developmental structure of lexical-semantic body knowledge in typically developing children, aged 4 to 12 years; and, 2) to explore the hypothesis regarding the influence of BS and BSD on the development of BI, using graph analysis to represent the associations among body parts. We are especially interested in somatosensorimotor influences on the development of lexical-semantic body knowledge. We hypothesize that BI knowledge is influenced by somatosensorimotor processes, and that knowledge of body parts develops continuously throughout the investigated age groups.

This investigation was carried out using graph analysis because graph structures represent the associations among elements and have been used to aid in the comprehension of complex systems in different areas of knowledge (Albert & Barabasi, 2002; Bullmore & Sporns, 2009). During the last decade, it was suggested that graph theory also presents a method for analyzing psycholinguistic tasks in healthy and clinical populations (Becker et al., 2014; Bertola et al., 2014; Lerner et al., 2009; Mota, et al., 2012; Zortea et al., 2014). This could provide a step in understanding the mechanisms underlying the development of body lexical-semantic knowledge related to the development of sensorimotor functioning. To date, a great number of studies have investigated only younger children, and no one has used a free naming of body parts task (word fluency) to investigate the development of BI.

Materials and methods

All research procedures complied with the Helsinki principles and were approved by the local ethics in research board (Research Ethics Committee of the Federal University of Minas Gerais). Informed consent was obtained in written form from parents or legal guardians and orally from children.

Participants

Two hundred fifty-one children between the ages of four and twelve years agreed to participate in the present study. They were recruited from public and private schools in Belo Horizonte and the surrounding metropolitan region (Minas Gerais, Brazil). All participants presented typical development, with no motor or language developmental delays reported by the parents or legal guardians. After evaluation, thirty-four children who performed below the fifteenth (15th) percentile in the intelligence task (Raven's Progressive Coloured Matrices - Angelini et al., 1999) and thirteen children who performed outside three standard deviations (extreme cases) in the word fluency task were excluded from the analyses. Therefore, the final sample comprised 204 individuals [mean age = 103 (sd = 31.5) months; 56.4% female]. The influence of sensorimotor processes on the development of semantic-lexical knowledge of the body parts was cross-sectionally investigated. Children were divided into three age groups: 4-6 years (n=69), 7-9 years (n=59), and 10-12 years (n=76). The three groups were homogeneous according to sex and intelligence ($p>0.05$). Descriptive data are shown in Table 1.

Instruments

General intelligence was evaluated using Raven's Progressive Coloured Matrices, validated for the Brazilian population (Angelini et al., 1999). Children presenting general intelligence below the fifteenth (15th) percentile were excluded.

Table 1 – Descriptive data of the sample.

	4-6 years		7-9 years		10-12 years					
	n	%	n	%	n	%	x^2	df	p	ϕ
Sex										
Male	29	14.2	30	14.7	30	14.7	1.86	2	0.39	0.09
Female	40	19.6	29	14.2	46	22.5				
	mean	**sd**	**mean**	**sd**	**mean**	**sd**	**F**	**df**	**p**	**η²**
Age (months)	63.85	9.22	106.78	12.30	134.80	11.83	503.76	2;201	<0.01	0.854
Raven (z-score)	0.55	0.89	0.62	0.68	0.54	0.65	204	2;201	0.81	0.002

The semantic word fluency task evaluates the spontaneous production of words under restricted search conditions (Strauss et al., 2006). The objective was that the child produce, as quickly as possible for 60 seconds, the largest number of examples within a semantic category. The semantic categories were: animals, foods and body parts. We compared the development of lexical-semantic body knowledge with two other categories of lexical-semantic knowledge (animals and foods), hypothesizing that only lexical-semantic knowledge of body parts should be influenced by bottom-up sensorimotor processes. All words produced by the participants were registered: total words, total correct words, total repeated words and total errors. For analysis in the present study, only correct and repeated words were considered. Children who scored below three standard deviations were considered extreme cases and excluded from the analyses.

Procedures

Data collection was conducted at the participants' schools, in two sessions of approximately 30 minutes each, by especially trained undergraduate psychology students. Intelligence assessments were applied to groups of approximately 6 children during the first session, and the semantic word fluency tasks (animals, foods and body parts categories) were individually assessed in the second session. The animals and foods categories served as controls for the body parts category in graph analyses.

Graph analyses

A graph is the mathematical representation of the relation between items, and this representation is expressed as a network (in this case, a semantic network) composed of a set of items, called nodes, and links between these items, called edges (Albert & Barabasi, 2002; Lerner et al., 2009; Mota et al., 2012). Each point corresponds to a node and, if two points have a relation to each other, they are linked by a line (Figure 1). The sequence of words produced in the semantic word fluency task was represented in an individual graph using the *SpeechGraphs software* (Mota et al., 2012).

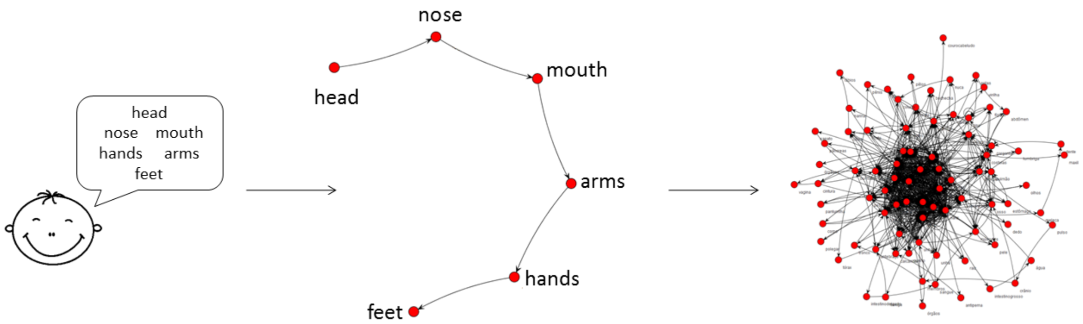

Figure 1 – Example of the representation, using graphs, of the sequence of words produced by 5 years old child. In sequence, example of a semantic network formed from the word fluency task by the children.

Using the words obtained in the semantic word fluency task, we calculated the sum of correct and repeated words (CRW). Five SpeechGraphs attributes (SGA) were also calculated. Those five attributes are the: nodes (N, number of words), edges (E, number of links connecting the nodes), density (D, number of edges divided by possible edges [D = 2*E/N*(N-1)]), diameter (DI, length between the node pairs of a network) and the average shortest path (ASP, average length of the shortest path between pairs of nodes of a network). It was expected that the children would produce graphs with N-1 edges and having low density (the larger the vocabulary, the larger the number of possible connections). As children develop, the number of nodes and edges increases as their vocabulary grows. However, the overall network structure should remain (edges = N-1). The networks should become less dense, due to the establishment of functional relations between the categories studied resulting in words organized in associative pairs.

Subsequently, to explore our hypothesis, graphs were created according to age for each category of words, forming semantic networks for each category studied. The words that composed the nuclei of the networks for each semantic category evaluated were investigated. The adoption of the nuclei of the semantic networks is based on the fact that the items most typical of a category are those produced with the greatest frequency. The resulting patterns of words were analyzed qualitatively.

Statistical analyses

Since the age groups were shown to be comparable, in relation to sex and intelligence, the parameters obtained from the graphs formed among the three groups in the word fluency task were compared using General Linear Models variance analysis (p-values < 0.05 were considered significant).

Results

As the evaluation of body knowledge relied on words representing body parts, it was possible that different word fluency of the children in each category could skew their responses. Thus, we measured the word fluency network metrics of each participant in all three categories. Table 2 shows that a similar age pattern for word fluency network metrics occurred across categories (foods, animals and body parts). This included a statistically significant increase ($p <0.001$) of CRW, nodes, edges, diameter and ASP among the children aged 7-9 years, that becomes more prominent in children aged 10-12 years when compared with children aged 4-6 years, for all semantic categories. A statistically significant ($p <0.001$) decrease of density was found among children aged 7-9 years when compared with children aged 4-6 years, for all semantic categories. However, we found a statistically significant ($p <0.001$) decrease of density only in the body parts category when we compared the children aged 7-9 years with those aged 10-12 years.

Analysis of word fluency network metrics yielded nine semantic networks, each depicting one word-category for each age group, as illustrated in Figure 2. The center of the network, the nucleus, comprises the words used most often, i.e., the core vocabulary of the children for the category, for that age group. Comparisons among age groups showed that the older group of children presented an increase of the number of nodes. So, it is possible to suggest that as children become older, the number of nodes (words) increases, resulting in growth in the size of the network nucleus.

Table 2 – Significant differences among groups in the three categories.

	4-6 years	7-9 years	10-12 years	F	df	p	η²
		mean (sd)					
Animals category							
CRW	10.15 (3.58)	13.55 (3.78)	15.96 (3.97)	42.473	2;201	<0.001*	0.297
N	9.39 (3.53)	12.83 (3.32)	15.58 (3.57)	56.836	2;201	<0.001*	0.361
E	9.06 (3.67)	12.37 (3.42)	15.11 (3.63)	51.340	2;201	<0.001*	0.338
D	0.27 (0.14)	0.17 (0.05)	0.14 (0.03)	42.255	2;201	<0.00+	0.296
DI	6.93 (3.50)	10.34 (3.76)	12.91 (4.69)	39.287	2;201	<0.001*	0.281
ASP	3.04 (1.13)	4.20 (1.23)	5.07 (1.45)	44.758	2;201	<0.001*	0.308
Foods category							
CRW	9.30 (3.23)	12.93 (3.77)	15.89 (4.18)	44.461	2;201	<0.001*	0.307
N	8.65 (2.88)	12.54 (3.53)	15.32 (4.75)	54.259	2;201	<0.001*	0.351
E	8.10 (3.11)	12.15 (3.76)	14.74 (4.85)	49.625	2;201	<0.001*	0.331
D	0.28 (0.12)	0.18 (0.06)	0.14 (0.04)	47.539	2;201	<0.001+	0.321
DI	6.65 (2.89)	10.20 (3.89)	13.00 (4.77)	46.426	2;201	<0.001*	0.316
ASP	2.91 (0.95)	4.12 (1,26)	5.04 (1.58)	47.784	2;201	<0.001*	0.322
Body parts category							
CRW	11.01 (3.60)	15.28 (3.89)	17.26 (4.59)	43.620	2;201	<0.001*	0.303
N	10.23 (3.07)	14.12 (3.60)	16.58 (4.14)	55.026	2;201	<0.001*	0.354
E	9.88 (3.44)	13.90 (3.85)	16.20 (4.45)	46.500	2;201	<0.001*	0.316
D	0.21 (0.07)	0.16 (0.05)	0.13 (0.03)	43.448	2;201	<0.001§	0.302
DI	7.80 (2.83)	10.93 (4.07)	13.29 (4.57)	35.452	2;201	<0.001*	0.261
ASP	3.29 (0.97)	4.39 (1.33)	5.19 (1.48)	39.158	2;201	<0.001*	0.280

*CRW, correct and repeated words; N, nodes; E, edges; D, density; DI, diameter; ASP, average shortest path.. Bonferroni Post-hoc: * 4-6 years < 7-9 years < 10-12 years; +4-6 years > 7-9 years = 10-12 years; §4-6 years > 7-9 years > 10-12 years.*

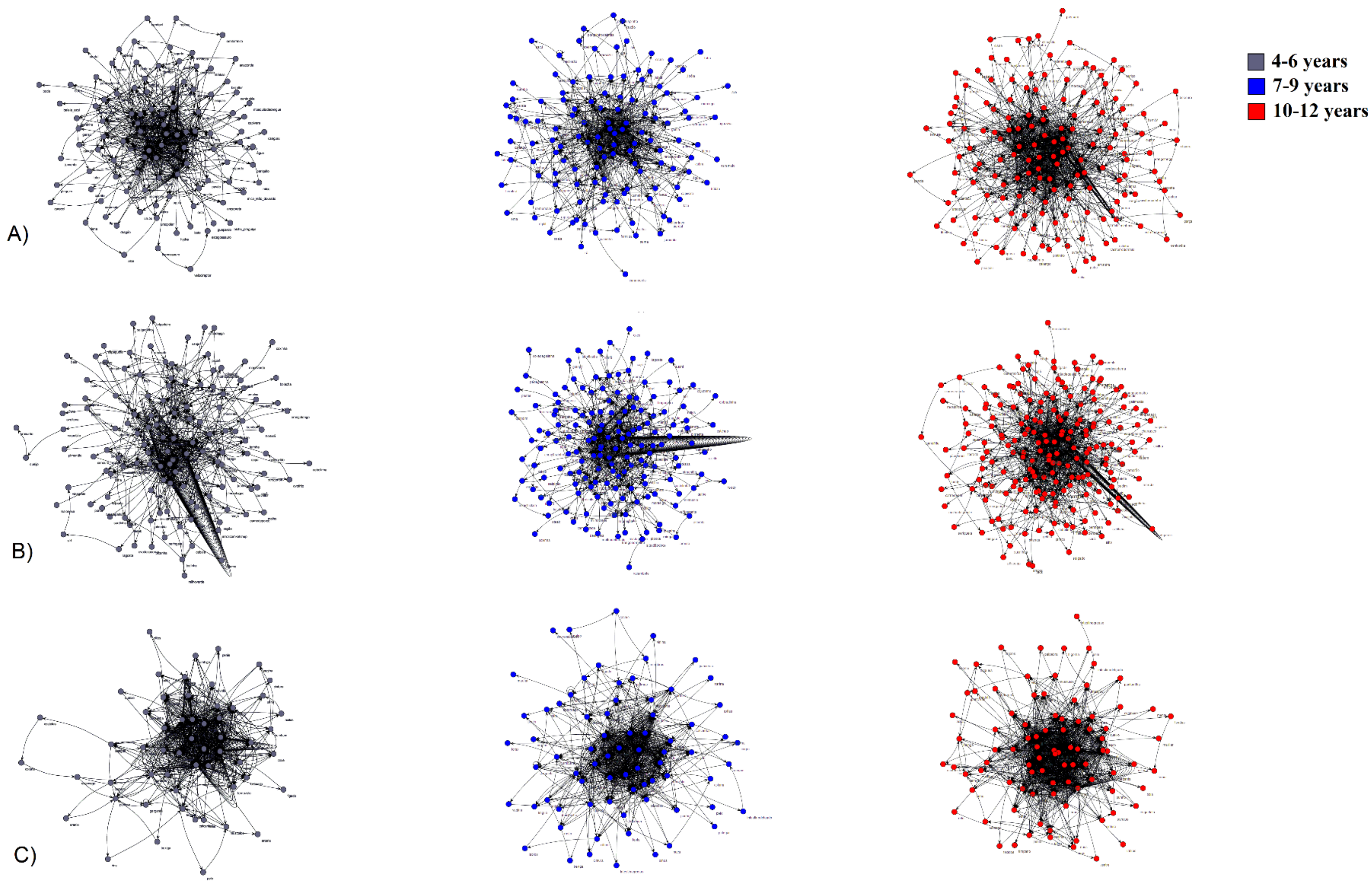

Figure 2 - Semantic networks formed from the children's word fluency tasks. A) animals category networks. B) foods category networks. C) body parts category networks. Each point in a network represents a word cited by children of that age group, and each line represents a semantic connection between the 2 words. Words closer to the center of the network are those used more often.

The concentric circles in Figure 3 indicate that as the ages of the groups increased, words were added to the nucleus of the network of the previous age group. This indicates a continuity of previously acquired vocabulary. However, when we tested whether word acquisition in each category clustered around distinct semantic features, no particular patterns emerged for the foods and animals categories. In contrast, for the body parts category, children aged 4-6 years mainly identified structures of the head and face, arms, hands, legs and feet; children aged 7-9 years identified the structures of the head and face, arms, hands, legs, feet and also the joints; and, children aged 10-12 years maintained the words used by the younger children and also identified the internal organs, sub-components of the limbs, and axial structures.

Discussion

To investigate how BI develops in children, we analyzed word fluency represented as networks in children aged from 4 to 12 years, subdivided into 3 age groups. The results can be summarized as follows: 1) the three groups of typically developing children performed distinctly in the three categories of the semantic word fluency task, with older children producing more words than younger children; 2) analysis of the properties of the semantic networks could distinguish typically developing children in different age groups; 3) semantic nuclei produced by the younger children were composed of common words retained in all older age groups; and, 4) analysis of the semantic network properties of the body parts category suggested an influence of infant sensorimotor development. This last result suggests that BS and BSD influence BI development, and will be discussed in more detail. Thus, the body parts word fluency networks indicated that the names for head/face structures and limbs typically are learned first, followed by the names for the joints and internal organs, implying that visual and somatosensorimotor development influences the body image. This hypothesis is reinforced by the fact that the acquisition of word fluency for the animals and foods categories, which have no relation to BI, followed no consistent pattern.

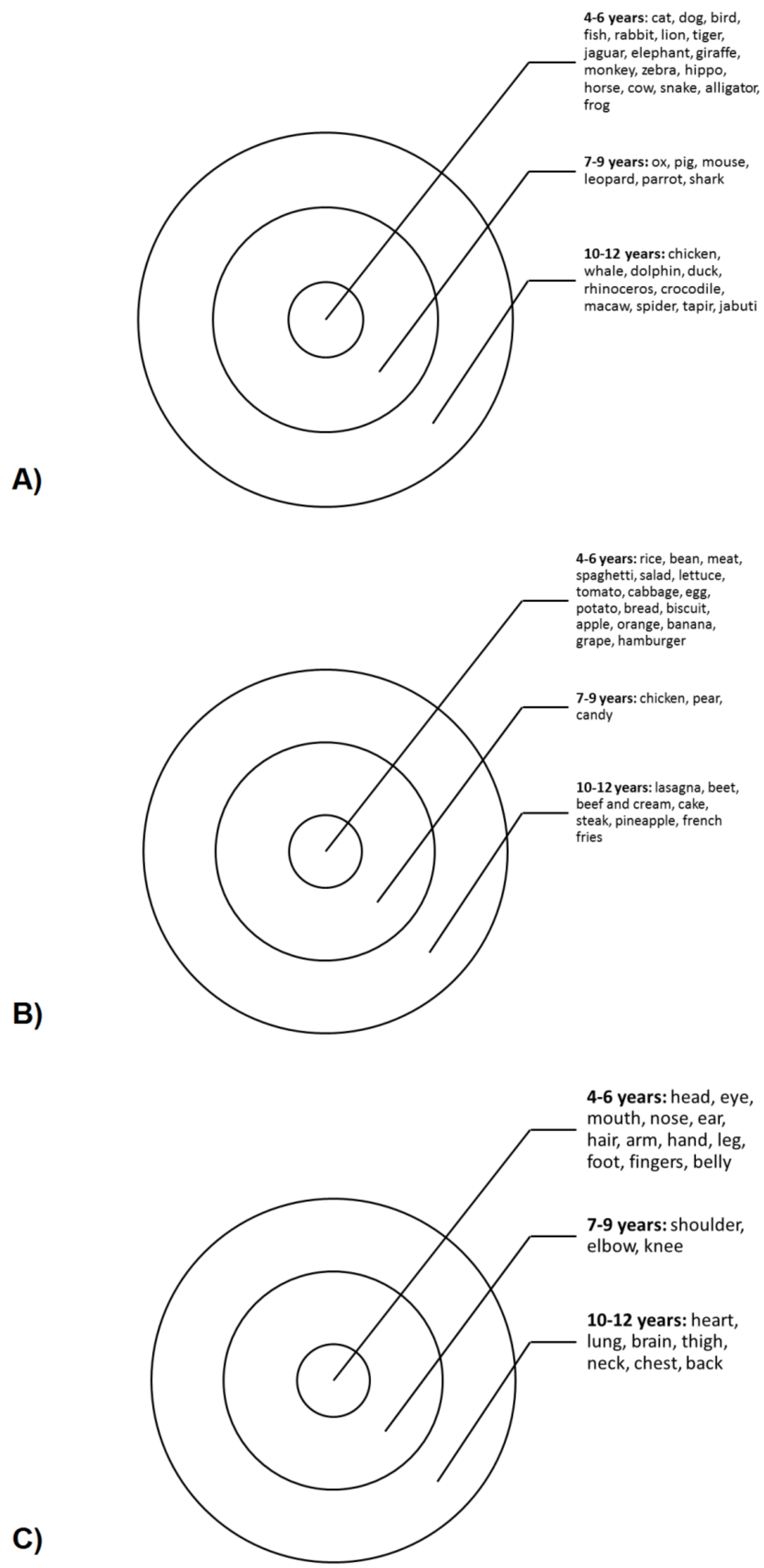

Figure 3 - The nuclei of the semantic networks illustrate continuity of semantic nuclei across the age groups. We found a possible developmental trend across the groups for the category "body parts". For the categories "animals" and "foods", no differences were observed. A) Nuclei of the animals category. B) Nuclei of the foods category. C) Nuclei of the body parts category.

Performance in the semantic word fluency task

The mean word fluency scores obtained by the children were similar to the normative data obtained in studies by Halperin et al. (1989), Malloy-Diniz et al. (2007), and Riva et al. (2000). Differences were found among the groups regarding the number of words produced in all semantic categories, in which younger children produced fewer words than older children. An increase in the number of words spoken by typically developing children throughout childhood is well established in the literature, and studies conducted in different countries show a clear improvement in age-related performance (Charchat-Fichman et al., 2011; Halperin et al., 1989; Malloy-Diniz et al., 2007; Riva et al., 2000; Sauzéon et al., 2004). The effect of age on categorical fluency performance tends to stabilize at around 11 to 12 years (Sauzéon et al., 2004).

One explanation for the changes in categorical fluency performance may be found in the progressive integration of the prefrontal cortex areas related to executive control and the temporal areas related to semantic knowledge (Jurado & Rosselli, 2007; Miyake & Friedman, 2012; Wright et al., 2015). This connection is hypothesized and supported by studies showing impairment in word fluency in individuals who have suffered injuries to the frontal areas and to the temporal lobe (Lopes et al., 2009; Rich et al., 1999; Tröster et al., 1998; Troyer et al., 1998).

Studies that have examined the role of connections among different cortical and subcortical brain structures in children also help us to understand this relationship. Research has shown that functional connectivity undergoes great changes, with a greater number of short-range brain connections being observed at the beginning of development. Throughout childhood and adolescence, functional connectivity becomes increasingly distributed, with long-range connections becoming stronger and short-range connections decreasing (di Martino et al., 2014; Fair et al., 2009; Rubia, 2013). Increases in age-related connections have been observed in studies with children aged from 6 to 10 years and from 7 to 18 years (Langen et al., 2018; Solé-padullés et al., 2015). Many of the significant, positive associations were identified in connections between regions in different lobes and/or hemispheres, and were in medium- to long-range connections (Langen et al., 2018). This increase in connections parallels the increase in the volume of the frontal, temporal and parietal lobes, which occurs in this same age group (Lenroot & Giedd, 2006). Increased volume can also result in increased cross-communication between brain regions (Langen et al., 2018). Studies using graph theory have also shown that, although the topological aspects of brain connectivity are mature at 8 years old, the modularity of brain networks continues to mature into adulthood (Menon, 2013).

Influence of age on structural characteristics of networks

The networks formed by the groups of children participating in the present study are conceptualized by Albert & Barabási (2002) as free-scale networks. These kinds of networks, unlike the networks represented by graphics in which the connections are random, have some nodes with many connections, and more nodes with fewer connections (Albert & Barabási, 2002). According to Lerner et al. (2009), the conservation of the graphic properties for the three age groups suggests that the basic mechanisms of categorical fluency are similar among groups. Thus, the exploratory analysis of the nuclei of the networks, of the typical performance of children in the semantic word fluency task in the present study, suggests no bias of general lexical access ability.

Differences in graph complexity are observed across age groups, characterized by the increased number of nodes and edges among the group aged 7-9 years when compared with the group aged 4-6 years. This difference is even more prominent in the group aged 10-12 years. This corroborates the finding that semantic access increases during children's development (Charchat-Fichman et al., 2011; Halperin et al., 1989; Malloy-Diniz et al., 2007; Riva et al., 2000; Sauzéon et al., 2004). These differences were also present in the global attributes diameter, density and average shorter path (ASP). These results indicate that the networks found for older children were more direct, had less repetition of words, thereby resulting in less dense networks. In addition to the increased vocabulary, it is possible to consider that older children have decreased network density due to the establishment of functional relationships among categories. If we consider the results obtained in the body parts category, they suggest a better understanding of the "self" with age. Thus, older children tend to produce words in more organized associative pairs (e.g., "foot-leg", followed by "hand-arm"), while younger children tend to quote words more randomly (while "head" may be stated by one child after "trunk", it also can be stated by another child after "knee").

The findings of Crowe & Prescott (2003) and Koren et al. (2005) corroborate this hypothesis by revealing an increase in the number of clusters formed by older children, suggesting continuity in the organization of concepts during children's development. Furthermore, specifically with regard to the body parts category, Auclair and Jambaqué (2014) observed that visuospatial representations of the body influence lexical-semantic processes. Thus, it may be suggested that organization of the elements of categories may derive from interaction between the individual and the environment.

Bottom-up theories suggest that categorization emerges from motor and sensory experiences. According to the sensory/functional theory (SFT) originally formulated by Warrington & McCarthy (1983; 1987) and Warrington & Shallice (1984), the semantic system: 1) is organized into semantic specific subsystems of modalities (e.g., visual/perceptual or functional/associative); and, 2) the ability to recognize/name living things depends on visual/perceptual information, while the ability to recognize/name artifacts depends on functional/associative information. Corroborating this theory, research on anatomical and clinical correlations in neuropsychological patients shows that, after injuries to the temporal neocortex in the ventral visual pathway, selective deficits are observed for the categories of living beings (Saffran & Schwartz, 1994, Gainotti et al., 1995). Parietal, frontal and temporal (dorsal visual pathway) injuries result in deficits related to artifacts categories (Saffran & Schwartz, 1994, Gainotti et al., 1995). Also, within the bottom-up or embodied cognition framework, studies suggest that conceptual processes are based on sensorimotor processes, supporting the notion that deficits for naming animals are associated with lesions of the anterior left ventral temporal cortex, and deficits for naming tools are associated with lesions in the posterior and lateral temporal areas (Damasio et al., 1996). In addition, deficits for naming tools and naming fruits and vegetables were associated with lesions to the inferior pre- and postcentral gyrus (Damasio et al., 2004).

Another point of view about category-specific semantic deficits is the domain-specific hypothesis (Caramazza & Shelton; 1998). This is a top-down theory, in which the authors assume that concepts are not directly related to sensorimotor experiences; rather, that they are represented outside of sensorimotor cortices and organized by conceptual properties, instead of perceptual properties (Caramazza & Shelton; 1998; Mahon & Caramazza; 2008). In a study with patients with optic aphasia, it was found that they could not name objects presented visually, but they were able to name the same objects when they were presented through the tactile modality. This indicates that the naming impairment is not due to an impairment in name retrieving (Hillis & Caramazza; 1995). Of course, semantic categories could originate from perceptual experiences and later dissociate from them.

The study by Huth et al. (2016), in which semantic selectivity across the cortex was mapped using functional MRI data, suggests that the organization of semantically selective brain areas is consistent across individuals. According to the authors, this might suggest that the organization of high-level semantic representations and their anatomical connection is innate or, at least, subject to extraordinarily similar experiences. It also could be a result of the lives of the subjects that participated in the study (all of whom grew up and were educated in the same region).

Similarity of semantic nuclei across groups

The structures of the semantic nuclei were qualitatively analyzed, considering differences in the graphic parameters across the groups. For all semantic categories, there was a common core for all age groups studied. With increasing age, words were added to the core network presented by the younger age group. The nucleus found for the younger age group was retained by the next age group.

A possible developmental trend in network structure across age groups was observed only for the nuclei in the "body parts" category. The body parts category presented a semantic network pattern suggesting a connection with somatosensorimotor development, due to the better performances in body parts related to action and sensory systems. This finding is consistent with the fact that the experience of the body as a unit and the continuity of the body itself depends on multisensory integration (Baumard & Osiurak; 2019).

On the other hand, for the "animals" and "foods" categories, no differences were observed in the nucleus structure at any age. According to Crowe & Prescott (2003), children tend to form groups according to their familiarity with the animals and not according classes (mammals, birds, fish, amphibians and reptiles) or habitats. Similar results were found by Lucariello, et al. (1992) and Grube & Hasselhorn (1996), showing that the animal groups are formed by the environmental context of the child. The same could probably happen with the foods category.

Also, the "body parts" most frequently identified in each age group were related to the children’s sensorimotor development. Thus, it can be assumed that the lack of differences among the age groups for "animals" and "foods" was due to the difference of sensory modalities involved in the semantic organization of the categories studied. For example, animals and foods are known and recognized for their visual/hearing and visual/taste characteristics, respectively. However, it is not possible to exclude a priori that semantic knowledge of pets is also influenced by kinetic/proprioceptive experience. Development of body parts knowledge seems to be influenced by proprioceptive sensations in addition to other sensory modalities. It is through the body itself that the individual interprets the stimuli offered by the external environment; and, there are extrinsic and intrinsic stimuli that modulate the knowledge and recognition of the body.

Interestingly, the body parts engaged in the recognition of characteristics of "animals" and "foods" (eyes, hands, ears, nose and mouth) are those with greater sensory representation and are identified beginning at the younger age. From a bottom-up perspective, as the sensory inputs involved were already well-developed representationally, these categories would not show a difference between the patterns in the age groups studied.

Camões-Costa et al. (2010) showed similar results with respect to the body parts most commonly identified and their presumed sensory representation in the cerebral cortex. Unlike other parts of the body which are rich in sensory afferents (facial and hand structures), joints (related to proprioceptive notions) were cited only from the second age group onward (7-9 years). This can be explained by the fact that this is a period characterized by consolidation and improvement of the basic movement patterns developed in early childhood (Eckert; 1993). Thus, refinement of basic motor patterns, adaptation of motor patterns to structural differences, improved coordination and motor control are characteristics of this age group.

Despite the fact that muscle spindles are mature in children as young as 3 years (Österlund et al., 2011), and that we examined children older than 4 years, studies reveal that threshold amplitudes for eliciting stretch and hoffman reflex responses do not reach adult levels until 6–7 years (Grosset et al., 2007; O’Sullivan et al., 1991). Based on elbow position matching studies, Goble (2010) suggests that children aged 8-10 years present an overall refinement of position matching ability that continues to develop into the adult years. Our data are consistent with data presented in developmental studies in which the joints are included in the semantic nuclei of body parts from the group aged 7-9 years suggesting an influence of the BS on BI. It could be observed in typically developing subjects that BI is highly dependent upon synchronous multimodal information (de Vignemont, 2010; de Vignemont, 2011). This seems to imply that development of body knowledge is based on visual and proprioceptive feedback (i.e., relies on BS).

The acquisition of lexical-semantic knowledge about the human body is an ongoing, lifelong process. Not only the body parts with visible and noticeable functions, such as the sensory organs of the face, hands, feet and joints, but the functions of the internal organs are also learned. The literature contains indications that functional knowledge of some internal organs of the body is acquired in preschool and early school years. From these ages, this knowledge develops progressively as children build a biological picture through formal learning about the physiology of the human body (Inagaki & Hatano, 2006; Jaakkola & Slaughter, 2002). This corroborates the results found in the present study, in which children identified the internal organs only in later childhood, when they had acquired knowledge of the human body.

Interaction between representational levels

Overall, the results of the development of the nuclei of the body parts corroborate findings from previous studies regarding learning about body parts (Auclair & Jambaqué et al., 2014; Camões-Costa et al., 2010; Crowe & Prescott, 2003; MacWhinney et al., 1987; Witt et al., 1990). The names for the structures of the head/face and limbs are typically learned first, followed by the names for the joints and internal organs. This trend in development is consistent with the development of the topographic representation of the body and body structure.

Simons and colleagues (Simons & Dedroog, 2009; Simons et al., 2011) found that children with intellectual disability and/or psychiatric disorders underperformed typically developing children in tasks that assess BSD and BI. Their results demonstrate the importance of topographic representation for lexical-semantic representation of the body.

Considering the motor correlates with sense of body, the Rubber Hand Illusion allows us to suggest that BSD overlaps with BS. In this set-up, subjects were asked to look at a prosthetic rubber hand positioned next to their own hand, which was hidden. In healthy controls, the illusion is created by synchronous tactile and visual stimulation (Botvinick & Cohen, 1998; Fotopoulou et al., 2008). In order to embody the rubber hand, it must first be integrated into the BS (Longo et al., 2009; Tsakiris & Haggard, 2005). The illusion also happened in patients with hemiplegia. However, patients with anosognosia were also unable to detect absence of movement correctly in trials where they, themselves, had to generate the movement (Fotopoulou et al., 2008). In this case, the hypothesis is that patients with anosognosia have difficulties with sensory feedback perception.

Impairments to sensorimotor functioning may have some impact on BI development. In a study involving individuals with schizophrenia, a reduced acuity in both BSD and BI tasks was observed, suggesting that the consequences of some alterations in BSD could conceivably exacerbate other body representation changes (Graham-Schmidt et al., 2016). An investigation of the consequences of a pediatric spinal trauma on body representation revealed a selective impairment in BI (Salvato et al., 2017). It was also suggested that body parts knowledge is related to sensorimotor experience.

Further studies are needed to confirm the hypothesis of the present study. The implication of sensorimotor influences on BI development was only inferred in the present study, as we did not specifically assess BS and BSD. Future studies should simultaneously assess the three levels of representation in order to investigate the possible interactions more directly. Longitudinal studies are also required to assess causal hypotheses.

Another way to better understand the impact of sensory and sensorimotor functioning and their interactions with body representation would be to evaluate the development of body representation in children with developmental disorders such as congenital blindness (Crollen et al., 2011; Crollen et al., 2014; Nava et al., 2014; Petkova et al., 2012) and cerebral palsy. It has been shown that children with cerebral palsy present disorders in the three levels of body representation (Fontes et al., 2014; Fontes et al., 2017; Souto et al., 2020). This raises some interesting questions. Is BI development impaired in cerebral palsy owing to disorders of sensorimotor processes? As the different levels of body representation interact, is it possible to compensate deficits in a more impaired level by stimulating the development of the relatively spared levels?

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4.3. Body experience influences lexical-semantic knowledge of body parts in children with hemiplegic cerebral palsy

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Abstract

Background: Disorders in different levels of body representation (i.e., body schema, body structural description, and body image) are present in hemiplegic cerebral palsy (HCP). However, it remains unclear whether the body image develops from aspects of body schema and body structural description, and how this occurs in children with HCP. Objective and methods: In a cross-sectional study, we investigated 53 children with HCP (mean age about 10 years) and 204 typically developing (TD) control children to qualitatively evaluate whether and how body schema (related sensorimotor experiences) and body structural description (related visuospatial experiences) affect the development of children's body image and whether this development is delayed through HCP. Graph analysis was used to create a lexical-semantic map of body representation from data of a semantic word fluency task. Results: Results indicated a similar qualitative pattern of influences of sensorimotor and visuospatial experiences on lexical-semantic knowledge of body parts, with a delayed developmental course in children with HCP compared to TD children. Conclusion: These findings suggest that children's body image seemed to be influenced by body schema and body structural descriptions as indicated by poorer lexical-semantic knowledge of body parts in children with HCP due to missing physical experiences of the affected body parts. This might imply that

"body talk" may beneficially complement physical therapy for children with HCP to promote body image development.

Key-words: body representation; body image; sensorimotor experience; neuropsychology; hemiplegic cerebral palsy

Introduction

Children with hemiplegic cerebral palsy (HCP) learn strategies to manage their everyday life using only one hand as the affected limb is usually being neglected or not used – a phenomenon known as developmental disregard (Houwink et al., 2013). They may present functional limitations related to the affected upper limb that cannot be explained by muscle strength impairments and may be aggravated by visuoperceptual disorders (de Ajuriaguerra & Stucki, 1969). Additionally, unilateral neglect may further impair the processing of perceptual information from the environment (Katz et al., 1998). Interestingly, children with HCP often also show atypical processing of information associated with their body, resulting in sensory deficits in their upper extremities such as threshold disturbances in proprioception as well as perception (Riquelme & Montoya, 2010). To account for these symptoms, the hypothesis of a disorder of higher-level body representation in HCP was proposed by de Ajuriaguerra & Stucki (1969) decades ago. Taken together, one might speculate that some symptoms observed in children with HCP may be due to an impairment of body representation at different levels (Fontes et al., 2014).

Body representations have been suggested to be organized into three neuropsychological levels: sensorimotor, visuospatial, and semantic-lexical (Golgenberg, 2002; Sirigu et al., 1991). The sensorimotor representation of the body, henceforth referred to as body schema (BS), incorporates proprioceptive information about the body itself and is characterized by continuous updating and consequent adaptation to changes in body properties and relative positions due to movements (Coslett et al., 2002; Golgenberg, 2002; Sirigu et al., 1991). The visuospatial representation, also termed and henceforth referred to as body structural description (BSD), describes the topographical representation of the body, providing information about its shape and surface contours as well as continuity and proximity relations among different body parts (Coslett et al., 2002; Golgenberg, 2002; Sirigu et al., 1991). Finally, body-related semantic-lexical knowledge is part of what we henceforth refer to as body image

(BI), which includes general information about names of body parts, associations of body parts with tools and artefacts, functions of different body parts, and affective information about the body (Coslett et al., 2002; Golgenberg, 2002; Sirigu et al., 1991).

In contrast to the extensive literature on representational deficits regarding the body in adults with (unilateral) brain damage, only few studies investigated impairments of body representation in brain-damaged children (Butti et al., 2019; Corti et al., 2018; Fontes et al., 2014; Fontes et al., 2017; Frassinetti et al., 2012). Of these, two examined all levels of body representation in children with HCP (Fontes et al., 2014; Fontes et al., 2017). Fontes et al. (2014) suggest that, similar to adult stroke patients, impairments of body representation in children with HCP are related to a decrease in spontaneous use of the affected limb not explained by motor problems directly associated with the respective brain damage. Fontes et al. (2017) reported evidence substantiating that damages to the immature brain, such as HCP, seem to drive disorders in body representation.

Impairments of different levels of body representation are dissociable in adults with brain damage (Sirigu et al., 1991; Schwoebel & Coslett, 2005), but nevertheless interact. The latter is inferred from the observation that BSD was observed to influence BS in experiments using the rubber hand illusion (Botvinick & Cohen, 1998). Moreover, in children aged between 5-10 years, BSD was observed to influence BI as indicated by children's naming performance for the location of the body parts (e.g., body parts vs. head features and also upper vs. lower limbs) or their involvement in motor skills (e.g., distal segments, joints, and broader body parts) (Auclair & Jambaqué, 2014). Furthermore, performance on tasks assessing BSD (e.g., finger gnosia, verbal and visual body parts localization, matching body parts by location) was found associated positively with performance on tasks measuring BS (e.g., imitation of meaningful and meaningless gestures) in a study investigating and comparing TD and children with HCP (Fontes et al., 2017). Also, performance on BI task (e.g., naming body parts) was associated positively with performance on tasks measuring BSD (e.g., finger gnosia, verbal and visual body parts localization, matching body parts by location) and BS tasks (e.g., hand laterality judgement task and imitation of meaningful gestures) (Fontes et al., 2017). Against the background of this brief overview of the literature, it seems that body representations develop in a more or less hierarchical manner with BSD gradually developing based on BS, and BI gradually developing from BSD.

However, little is known so far about whether and how body representations of children with HCP develop as compared to typically developing (TD) children. In particular, there are only few investigations of how an injury to the immature brain may impact the development of body representations during childhood (Christie & Slaughter, 2009; Simons & Dedroog, 2009; Simons et al., 2011). Therefore, this study investigated the development of BI using a word fluency task comparing the performance of TD children with that of children with HCP. We were particularly interested in whether BS (related sensorimotor information) and BSD (related visuospatial information) contribute to the development of BI (by qualitatively analyzing the body parts most cited in the word fluency task), and whether this development is delayed in children with HCP. As such, we compared performance on word fluency not only for body parts but also for animals, based on lexical-semantic maps using Graph Analysis across different age groups and comparing TD children and children with HCP.

Methods

Participants

This cross-sectional study involved a convenience sample of children with a diagnosis of HCP recruited in rehabilitation centers in Minas Gerais (Brazil). TD control children were recruited from public and private schools in Minas Gerais (Brazil). Children eligible to participate in the study met the following inclusion criteria: i) performance above 15th percentile in assessment of general cognitive ability, ii) no uncontrolled epilepsy and iii) ability to respond to the assessment procedures. The sample comprised 257 children in total, of which 204 were TD control children (age range 4-12 years, mean age = 8.09 years, SD = 2.60 years) and another 53 children with HCP [age range 7-12 years; mean age = 10.19 years, SD = 1.83 years; 36 right hemiplegic cerebral palsy (RHCP) and 17 left hemiplegic cerebral palsy (LHCP)]. To evaluate a potential delay in BI development, we compared performance of children with HCP to that of TD children separated into three age groups: i) 4-6 years (n = 69; mean age = 5.40 years, SD = 0.72 years), ii) 7-9 years (n = 59; mean age = 8.89 years, SD = 1.03 years), and iii) TD 10-12 years (n = 76; mean age = 11.21 years, SD = 0.96 years).

Ethics

This study was approved by the Research Ethics Committee of the Federal University of Minas Gerais (protocol number 2.155.379). Participation was conditioned to written informed consent from parents or legal guardians, and oral consent from children.

Materials

General cognitive abilities

General cognitive abilities were assessed using the Raven's Progressive Coloured Matrices (RCPM – Angelini et al., 1999) validated for the Brazilian population. Children who scored below the 15^{th} percentile were not considered for the study. Analyses considered z-scores (M = 0, SD = 1), computed as described in the test manual.

Semantic Word Fluency Task

The Semantic Word Fluency task evaluates the spontaneous production of words under restricted search conditions (Strauss et al., 2006). In two runs, each child had to produce as many animals in one and body parts in the other run, respectively, within 60 seconds each. We recorded the total number of words produced, total number of categorically correct words produced, total number of repetitions, and total number of intrusion errors as measures of children's performance. The number of categorically correct and repeated words was considered as dependent variable in the graph analysis.

Procedure

Data collection took place in schools and rehabilitation centers that children attended. Assessment of general cognitive abilities and application of the Semantic Word Fluency task were carried out by a team of trained undergraduate students in one-on-one sessions lasting about 40 minutes per child.

Graph Analysis

The sequence of words produced in the Semantic Word Fluency task was represented as an individual graph using *SpeechGraphs* software (Mota et al., 2012). The graphical structure reflects associations between a set of items expressed in the form of a network composed of nodes and edges, where nodes represent the items (i.e., words produced by children) and the edges the connections between these items (Albert & Barabasi, 2002; Mota et al., 2012). In addition to the sum of categorically correct words as well as repetitions (number of correct words and number of repetitions - CWR) obtained from the verbal fluency task, the software estimated six attributes: i) number of nodes (N); ii) number of edges (E); iii) density (D - number of edges divided by the number of possible edges), iv) diameter (DI), and v) average shortest path, (ASP - the shortest path length between pairs of more distant nodes in a network) (Mota et al., 2012). Better semantic networks would be indicated by N-1 edges of low density and with great distances, thereby generating direct graphs. When words were repeated, the graphs generated present $E \geq N$ and high density. In addition to individual graphs, group graphs were created to reflect semantic networks of children with HCP and the three age groups of TD children. Semantic network scores for body parts were used to identify the most frequent or typical words, which were then used for further analyses.

Statistical analyses

Preliminary analyses indicated that children with LHCP and RHCP did not differ in their scores on general cognitive ability as well as the semantic word fluency. Therefore, these two groups were pooled for the analyses.

In a next step, analyses of variance (ANOVA) were conducted to evaluate differences in general cognitive abilities between the group of children with HCP and the three different age groups of TD children. Despite scoring above percentile 15, the ANOVA revealed a significant effect of participant group on children's scores for general cognitive ability ($F_{(4;256)}=7.945; p < 0.01; \eta^2_p = 0.11$). Bonferroni corrected pair-wise comparisons indicated that children from the HCP group (M = -0.03, SD = 0.49) had significantly lower scores than the three TD groups (*all* $p < 0.001$; TD 4-6 years: M = 0.55, SD = 0.89; TD 7-9 years: M = 0.62, SD = 0.68; and TD 10-12 years: M = 0.54, SD = 0.64), whereas there was no significant difference between the three groups of TD children (*all* $p > 0.05$). Therefore, we considered general cognitive ability as a control variable in our subsequent analyses.

For the Semantic Word Fluency task, group differences in the number of correct words, repeated words and errors, as well as parameters obtained from the graph analysis, were analysed using mixed model analyses of covariance (ANCOVA) discerning the between-participant factor group (i.e., children with HCP vs. the three different age groups of TD children) and stimulus category (i.e., animals vs. body parts) while controlling for influences of general cognitive abilities. Additionally, we evaluated performance in the word fluency task using within-participant repeated measures ANOVA discerning the number of correct animals and number of correct body parts for each participant group.

We also explored the effects of BS and BSD on BI by qualitatively analysing words that composed the semantic network nuclei for the four groups.

Results

Semantic word fluency task

Number of categorically correct words produced

The mixed model ANCOVA revealed a significant main effect of group for the number of correct answers. Table 1 provides statistical details and descriptive results. Post-hoc pairwise comparisons indicated that there was no significant difference between children with HCP and TD 4-6 years for both animals and body parts ($p>0.45$). Children from the TD 7-9 and TD 10-12 groups produced more animals and body parts than children with HCP and those from the TD 4-6 group (all $p<0.001$). Finally, children from the TD 10-12 group produced more animals and body parts than children from the TD 7-9 group ($p<0.001$).

Additionally, the main effect of stimulus category was significant indicating that overall children produced more body parts than animals within the respective 60 seconds runs (Table 2). Interestingly, simple effects for the individual groups indicated that this was only the case for all TD control groups (all $p<0.02$), but not for children with HCP ($p=0.13$). Additional Baysian analysis following the recommendations by Masson (2011) of the posterior probability substantiated that there was no difference between the number of animals and body parts produced by children with HCP (>0.63 probability) by providing weak evidence in favor of the null hypothesis. The interaction of group and stimulus category was not significant though.

Table 1. Results of the semantic word fluency task. Comparisons between typically developing children group (TD groups: TD 4-6 years, TD 7-8 years, TD 10-12 years) and children with hemiplegic cerebral palsy (HCP).

	TD 4-6 years	TD 7-9 years	TD 10-12 years	HCP	F (3;252)	p	Partial η2	Post-hoc (Bonferroni test)
	mean (sd)							
Animals								
Correct words	9.22 (3.31)	12.81 (3.36)	15.38 (3.74)	9.94 (2.82)	46.726	<0.01	0.357	HCP = TD 4-6 years < TD 7-9 years < TD 10-12 years.
Repetitions	0.94 (1.40)	0.75 (1.35)	0.58 (1.36)	0.49 (0.75)	2.164	<0.09	0.025	-
Errors	0.16 (0.47)	0.05 (0.22)	0.01 (0.11)	0.04 (0.19)	3.359	<0.01	0.038	HCP = TD 4-6 years = TD 7-9 years; HCP = TD 7-9 years = TD 10-12 years; TD 4-6 years > TD 10-12 years.
Body parts								
Correct words	10.26 (3.31)	14.15 (3.45)	16.61 (4.40)	10.47 (3.52)	43.288	<0.01	0.340	HCP = TD 4-6 years < TD 7-9 years < TD 10-12 years.
Repetitions	0.75 (1.02)	1.14 (1.49)	0.66 (1.09)	0.60 (0.86)	2.784	<0.08	0.032	-
Errors	0.49 (1.14)	0.17 (0.37)	0.07 (0.25)	0.06 (0.23)	7.198	<0.01	0.079	TD 4-6 years > TD 7-9 years = TD 10-12 years = HCP.

TD 4-6 years = Typically developing children group (4-6 years old); TD 7-9 years = Typically developing children group (7-9 years old); TD 10-12 years = Typically developing children group (10-12 years old); HCP = hemiplegic cerebral palsy; sd = standard deviation; F = ANCOVA's ratio F; partial η2 = partial eta squared.

Table 2. Comparison between animals and body parts production (correct words).

Groups	Animals	Body parts	F	p	Partial η^2
	mean (sd)				
HCP	9.94 (2.82)	10.47 (3.52)	2.308	<0.135	0.043
TD 4-6	9.22 (3.31)	10.26 (3.31)	5.935	<0.017	0.080
TD 7-9	12.81 (3.36)	14.15 (3.45)	9.251	<0.004	0.138
TD 10-12	15.38 (3.74)	16.61 (4.40)	6.317	<0.014	0.078

TD 4-6 years = Typically developing children group (4-6 years old); TD 7-9 years = Typically developing children group (7-9 years old); TD 10-12 years = Typically developing children group (10-12 years old); HCP = hemiplegic cerebral palsy; sd = standard deviation; F = ANCOVA's ratio F; partial η2 = partial eta squared.

Finally, the covariate significantly influenced the results for the number of correct answers for both animals ($p<0.02$) and body parts ($p<0.01$) with children with higher general cognitive ability producing more correct answers.

Repetitions

There was no significant difference neither between groups nor for stimulus category for the number of repetitions with the respective main effects being not significant. Additionally, the interaction was also not significant. Covariate was not significant for the number of repetitions for both animals ($p>0.06$) and body parts ($p>0.51$).

Number of errors committed

There was a significant main effect of group for errors committed. Post-hoc pairwise comparisons indicated that there was no significant difference between the number of errors committed by children with HCP and children in the TD 7-9 years and 10-12 years age groups for both animals as well as body parts (all $p>1.00$). The number of errors committed by children with HCP and children in the 4-6 years for animals categories was not significant ($p>0.29$). However, the number of errors committed by children with HCP was significantly lower than the number of errors committed by TD children in the 4-6 years group for body parts categories ($p<0.001$). The number of errors committed by TD children in the 4-6 years group was significantly higher than the number of errors committed by TD children in the 10-12 years group for animals category ($p<0.04$), and higher than the number of errors committed by TD children in the 7-9 TD 7-9 years and 10-12 years age groups (all $p<0.03$). There was no significant difference for the number of errors committed by TD 7-9 years and 10-12 years age groups ($p>1.00$) . The interaction was not significant. Covariate was also not significant for the number of repetitions for both animals ($p>0.4$) and body parts ($p>0.20$).

Graph parameters

The mixed model ANCOVA revealed significant main effect of group for all parameters (see Table 3). Bonferroni-corrected pairwise comparisons indicated the three TD groups differed significantly from each other with respect to the number of nodes, density, diameter and mean of the shortest path (all $p<0.03$). For children of the TD 10-12 years group graphs were significantly less dense with a higher number of nodes and edges, larger diameters and ASP than for children of the TD 7-9 years and TD 4-6 years groups. The group TD 7-9 years presented intermediate parameters, which differed significantly from all parameters presented in the other TD age groups. The graphical parameters obtained for the HCP group differed significantly from the parameters obtained for the TD 7-9 years and TD 10-12 years groups (all $p<0.001$), but showed no significant difference to parameters observed for the TD 4-6 years group. The interaction was not significant. Covariate was also not significant for the graph parameters (all $p>0.7$).

To substantiate the observed null effect for the differences between the children with HCP and those from the TD 4-6 year group, we again conducted Bayesian analysis as recommended by Masson (2011). The comparison of the HCP group with the TD 4-6 years group revealed >0.89 probability and thus positive evidence in favor of the null hypothesis (Table 4) according to classification guidelines proposed by Masson (2011).

Semantic Network Cores

The networks formed by HCP group and TD 4-6 years, TD 7-9 years, and TD 10-12 years groups and the semantic nuclei obtained from the networks, which represent the words quoted more frequently for each category, are shown in Figures 1 and 2. All groups presented a common central semantic network core.

Table 3. Comparisons among groups in word fluency task (graph analysis of body parts category).

	TD 4-6 Years	TD 7-9 years	TD 10-12 years	HCP group	F (3;252)	p	Partial η²	Post-hoc (Bonferroni test)
	mean (sd)							
Nodes	10.23 (3.07)	14.12 (3.60)	16.58 (4.14)	10.13 (3.34)	48.792	<0.001	0.367	*HCP = TD 4-6 years < TD 7-9 years < TD 10-12 years*
Edges	9.88 (3.44)	13.90 (3.85)	16.20 (4.45)	9.72 (3.62)	41.663	<0.001	0.332	*HCP = TD 4-6 years < TD 7-9 years < TD 10-12 years*
Density	0.21 (0.07)	0.16 (0.05)	0.13 (0.03)	0.23 (0.08)	33.493	<0.001	0.285	*HCP = TD 4-6 years > TD 7-9 years > TD 10-12 years*
Diameter	7.80 (2.83)	10.93 (4.07)	13.29 (4.57)	7.53 (2.81)	34.643	<0.001	0.292	*HCP = TD 4-6 years < TD 7-9 years < TD 10-12 years*
Average Shortest Path	3.29 (0.97)	4.39 (1.33)	5.19 (1.48)	3.22 (0.94)	37.861	<0.001	0.311	*HCP = TD 4-6 years < TD 7-9 years < TD 10-12 years*

TD 4-6 years = Typically developing children group (4-6 years old); TD 7-9 years = Typically developing children group (7-9 years old); TD 10-12 years = Typically developing children group (10-12 years old); HCP group = hemiplegic cerebral palsy group; sd = standard deviation; F = ANCOVA's ratio F; partial η2 = partial eta squared.

Table 4. Bayesian analysis investigating non-significant differences between the HCP and TD 4-6 year groups.

Graph parameter	**HCP**	**TD 4-6 years**	***df***	SS_{effect}	SS_{error}	***F***	***BF***	$p_{BIC}(H_0\|D)$
Nodes	10.13 (3.34)	10.23 (3.07)	1; 119	4.516	1175.270	0.457	8.68302409	0.89672648
Edges	9.72 (3.62)	9.88 (3.44)	1; 119	3.816	1433.889	0.317	9.3131287	0.90303621
Density	0.23 (0.08)	0.21 (0.07)	1; 119	0.001	0.741	0.131	10.0675661	0.9096459
Diameter	7.53 (2.81)	7.80 (2.83)	1; 119	0.897	940.861	0.113	10.3074047	0.91156238
Average Shortest Path	3.22 (0.94)	3.29 (0.97)	1; 119	0.131	106.801	0.146	10.1413887	0.91024458

SSeffect = sum of squares for the effect; SSerror =sum of squares for errors; F = ANCOVA's ratio F; BF = Bayes factor; $p_{BIC}(H_0|D)$ *= posterior probability generated by bayesian information criterion (BIC).*

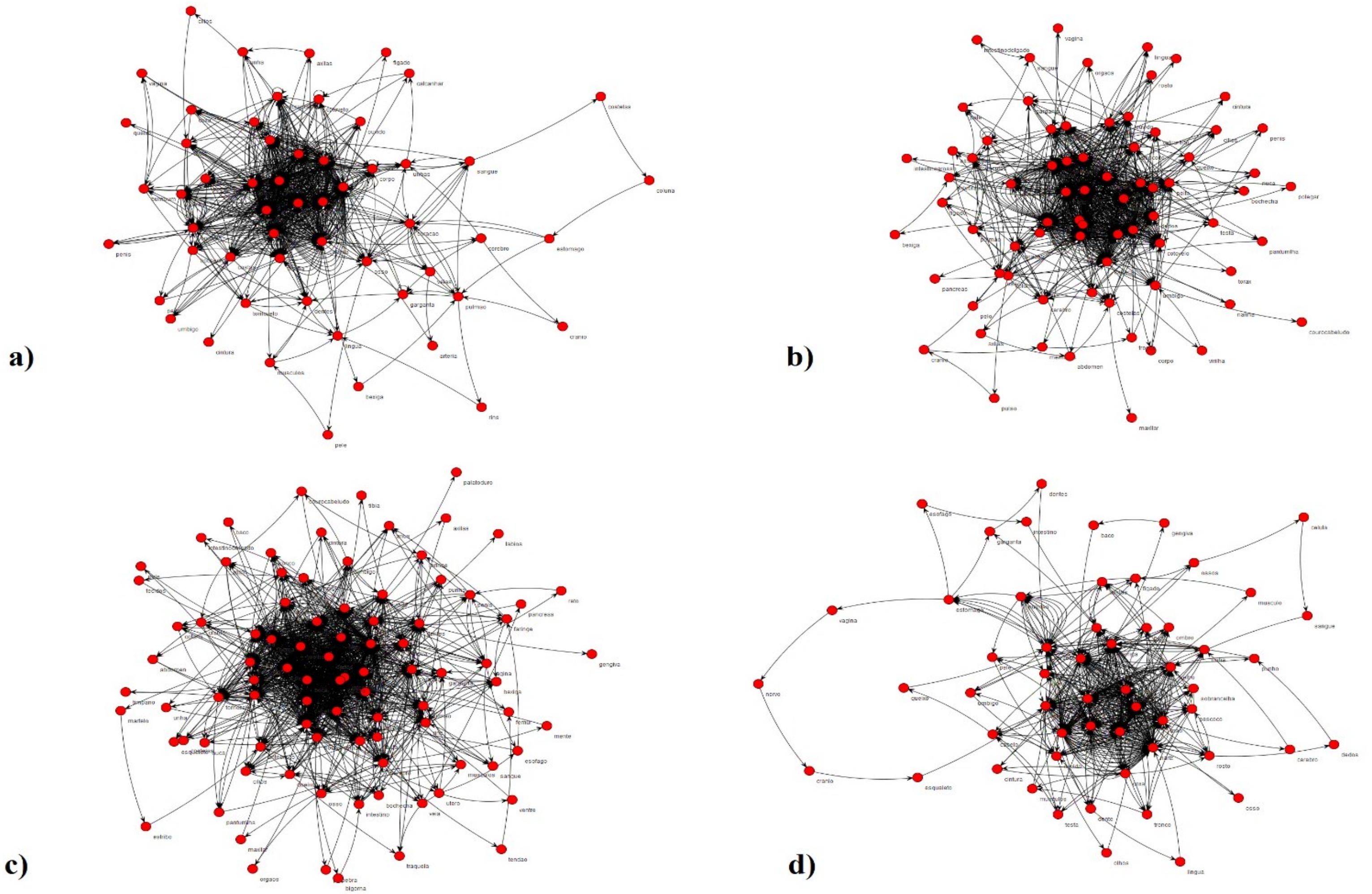

Figure 1. Semantic networks of body parts category formed by the groups. In a) semantic network formed by TD 4-6 years; b) semantic network formed by TD 7-9 years; c) semantic network formed by TD 10-12 years; d) semantic network formed by HCP group.

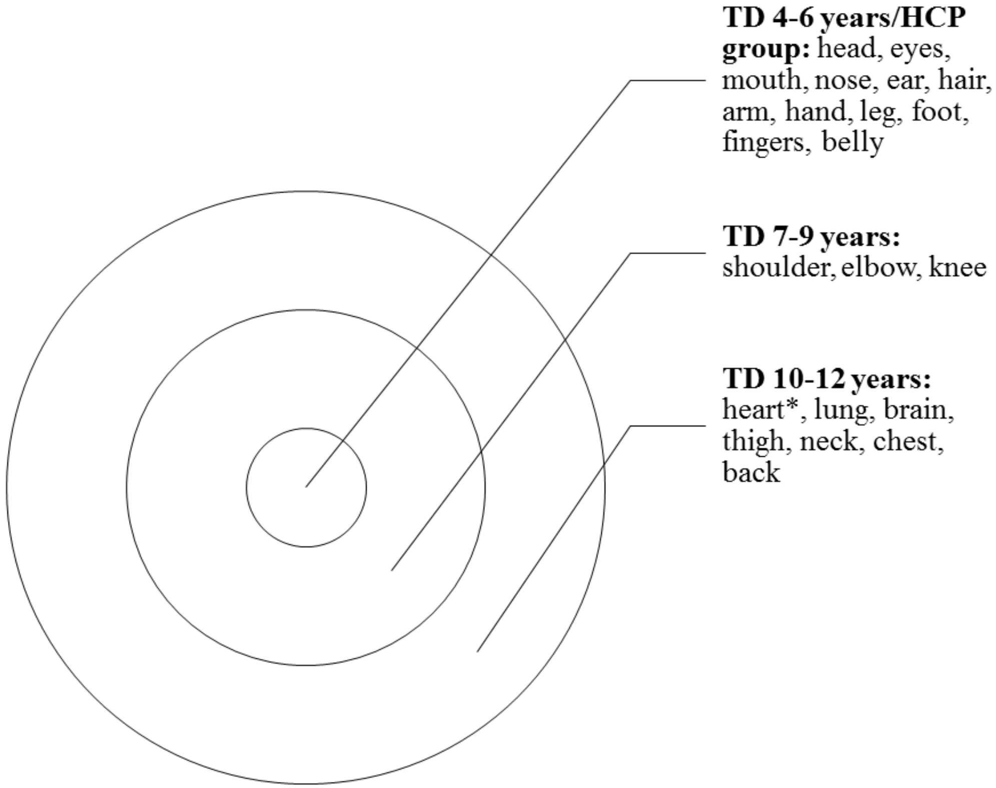

Figure 2. Illustration of the semantic network core obtained from networks, representing the words quoted more frequently for each category. All groups presented a common central core. *TD 4-6 years = Typically developing children group (4-6 years old); TD 7-9 years = Typically developing children group (7-9 years old); TD 10-12 years = Typically developing children group (10-12 years old); HCP group = hemiplegic cerebral palsy group; *heart was also cited by HCP group.*

Discussion

The aim of the current study was to investigate the development of BI using a word fluency task and compare performance of TD on this task to performance of children with HCP. We not only evaluated performance on word fluency for body parts but also for animals, based on lexical-semantic maps of the BI generated with Graph Analysis across different age groups and between TD and children with HCP. Apart from quantitative differences between groups and stimulus categories, we were also interested in examining (by qualitatively analyzing the body parts most cited) whether BS (related sensorimotor information) and BSD (related visuospatial information) contribute to the development of BI, and whether this development is delayed in HCP.

Children with HCP presented a representational profile of BI (as reflected by their performance in the semantic fluency task), which seemed equivalent to that of children from the TD 4-6 years group. However, they performed significantly worse than TD children of the other age groups including those of the same age. This may reflect a continuing maturation of BI in TD children not seen in children with HCP in a comparable manner. These results will be discussed in more detail in the following.

Semantic word fluency in HCP children

Children with HCP (aged from 7 to 12 years old) performed significantly worse than TD children 7-9 years and 10-12 years of age as regards the number of correct words produced, related to retrieval of the semantic memory content. This is consistent with previous findings (Carlsson et al., 1994; Kolk & Talvik, 2000). Interestingly, they performed comparably to the youngest TD group (i.e., 4-6 years of age) as substantiated by Bayesian analyses. In relation to the number of errors and repetitions in semantic word fluency (reflecting influences of executive functions - Anderson, 2002), children with HCP did not perform significantly different than TD 7-9 years and TD 10-12 years. This contrasts with previous research, which observed an impairment of executive functions (evaluated by verbal fluency) following early brain injury (Bodimeade et al., 2013). In addition, there was no evidence for differences in semantic verbal fluency according to side of hemiplegia (Bodimeade et al., 2013). According to a recent review, results regarding the presence of language impairments in children with HCP are inconclusive and whether they are observed might be due to differences in neural reorganization, and in location and extent of neural lesions (Bottcher, 2010; Liegeois et al., 2004).

Regarding potential differential influence of HCP on verbal fluency for animals and body parts, our results indicated a significant difference in the number of body parts and animals produced, with more body parts than animals produced overall. However, this difference was only significant for the control groups (TD 4-6, TD 7-9, and TD 10-12 years). For the HCP group this advantage for body parts was not observed. This may reflect a specific relative impairment for the representation of body parts due to HCP.

Structural characteristics of lexical-semantic body representation networks

Graph-theoretical analyses revealed qualitatively similar profiles for children with HCP and TD children. The qualitative conservation of the basic graphical properties across the four groups suggests that basic mechanisms of categorical fluency might be similar (Albert & Barabasi, 2002; Vitevitch, 2008). This also implies that connections formed may not be random, because some nodes presented many connections and many more nodes had few only connections, characterizing a free scale network. Free scale networks emerge from growth and preferential attachment mechanisms. Growth refers to the addition of new nodes (reflecting words cited) to the network over time (Vitevitch, 2008). Preferential attachment is a constraint that makes it more likely for new nodes being added to the system to connect to nodes that are already highly connected (Vitevitch, 2008). In terms of words, it means that a new word included in the networks will be probably connected to the words that were produced more often previously.

Overall, performance of children with HCP was quite similar to that of the group TD 4-6 years with quantitative parameters suggesting a lower degree of complexity of their networks than those presented by TD children older than seven years. Also, the semantic networks produced by TD 7-9 years and TD 10-12 years groups were more direct (with less repetition of words), resulting in less dense networks. In addition to the larger vocabulary of older children their networks probably also reflect the establishment of functional relations between body parts (e.g., feet are named after legs, or hands after arms). In contrast, children with HCP performed similar to the TD 4-6 years group and thus the youngest group of control children at the beginning of their BI development. This finding might reflect differences in sensory experience between children with HCP and their TD peers, as discussed below.

Sensory experience and lexical-semantic body representation in HCP children

Our data on the semantic networks for body parts in TD children suggest a developmental pattern similar to that observed previously in studies of body part identification (Auclair & Jambaqué, 2014; Camões-Costa et al., 2010; Christie & Slaughter, 2009; Witt et al., 1990). In all groups of TD children, words denoting specific body part categories (e.g., face structures, limbs, joints, internal organs) were added to the semantic network cores as age increased. Children of the group TD 4-6 years were found to primarily produce head / face structures and limbs in a non-hierarchical way (including arms, hands, legs, and feet but not dividing the upper limb into shoulder, arm, elbow, forearm, wrist, etc.). This might reflect influences from sensorimotor afferences contributing to BS (Christie & Slaughter, 2009). Parts of the body that receive more pronounced and early sensorimotor inputs (such as hands) may be learned preferentially (Ayres, 1961). This is substantiated by correlational analyses indicating that the body parts most frequently named by children are the structures best represented in the sensory cortex (Camões-Costa et al., 2010).

Joints were first mentioned systematically by children of the TD 7-9 years group. When reaching seven years of age, children are in a period of consolidation and improvement of the basic patterns of movement developed as compared to early childhood (Goodway et al., 2019). In this age group, a refinement of basic motor patterns, adaptation of motor patterns, improvement of coordination, and motor control is observed. These new sensorimotor experiences depend on tactile, kinesthetic, proprioceptive, vestibular and visual inputs. According to this line of reasoning, somatosensory afferences underlying BS may also influence the development of BI at this age, improving the ability to identify and name body parts.

Only at 10-12 years did the children add internal organs and hierarchize the limbs (e.g., arm and forearm, etc.) and axial structures (e.g., neck, nape, trunk, belly, etc.). Visuospatial experience contributing to BSD seemed to influence representations of BI at this age (Auclair & Jambaqué, 2014). Following this rationale, it seems possible that internal organs might only be learned later because they are not visible. The most salient and visible parts of the body are more identifiable and have easily observable functions and may therefore be learned before other non-visible and harder to experience parts of the body. Functional knowledge of some internal body parts may also emerge from formal learning about the biology of the human body (Auclair & Jambaqué, 2014; Christie & Slaughter, 2009).

The hierarchy of some axial structures (such as the division of the trunk into the neck, chest and back) only occurs later in development. This may be due to the influence of motor learning about joints and cultural influences related to formal learning about the human body (Jaakkola & Slaughter, 2002). Studies suggest that body parts can be segmented (e.g., the arm might be considered in whole as the superior limb, or the body part jointed to the forearm by the elbow) according to language, and the division of body parts can vary between different languages (Enfield, 2006; Majid, 2010). Older children are more experienced and more likely to expand their vocabulary, and the development of language is very closely related to the development of body awareness (Facon et al., 2002). Despite this, children with HCP (aged from 7 to 12 years old) presented a lexical-semantic network of body parts similar to that of the youngest TD 4-6 years group (as substantiated by Bayesian analysis). This is in line with but also expands previous studies which suggested that children with unilateral brain injury present lower performance in pointing (BSD) and naming (BI) body-part tasks than TD children (Christie & Slaughter, 2009; Fontes et al., 2017).

Although joints are expected to be a part of the semantic network core of children with HCP because they were part of the semantic-lexical repertoire of children of the same age, we did not observe these children to name joints in the semantic fluency task. This is an important aspect because joints are a point of reference for the segmentation of body parts, representing more detailed knowledge about the structuring of the human body (de Vignemont et al., 2009). Segmentation of the body into parts may derive from the organization of the proprioceptive and motor systems, or from perceptual factors such as the visual discontinuity of the body parts (de Vignemont et al., 2009). Following this rationale, motor activity may help to structure the mental representations of the body into functional units, according to the parts of the body that move together. In addition to representing anatomical points of reference, joints constitute the kinesiological basis of movement because the brain needs to identify the joints' position (from a set of proprioceptive signals coming from muscles, tendons, ligaments and joint capsule) and then plan the desired motor action (Marini et al., 2018). Difficulties in controlling movements, as often experienced by children with HCP, may thus influence their functional performance by restricting new sensorimotor experiences.

For effective motor action, for instance, when manipulating objects, it is necessary to represent the positioning and configuration of the upper limb to avoid uncomfortable or movement restrictive postures (Mutsaarts et al., 2006). Planning impairments have also been reported in young adolescents with HCP (Mutsaarts et al., 2006; Souto et al., 2020). Our study points to a delay in the development of lexical-semantic knowledge of body parts in children with HCP, which might reflect reduced sensorimotor and visuoperceptual experiences of their own body. Thus, it is plausible that lexical-semantic knowledge of body parts is influenced in a bottom-up manner.

When interpreting the results of the current study, some limitations have to be considered. The group of children with HCP was rather small, making it impossible to create subgroups of different ages for this group. Moreover, future studies might also include tasks to evaluate BS and BSD and measures for other executive functions components. We used a controlled word fluency task to assess children's knowledge on body parts. This test has a considerable higher degree of freedom compared to responses in a task requiring the naming of body parts.

Furthermore, graph-theoretical analysis identified an emergent structure of lexical-semantic network, qualitatively similar but less complex in children with HCP compared to TD children. Our results also suggested that building of the lexical-semantic network for body parts and thus BI seems influenced by sensorimotor and visuoperceptual experiences. As suggested by Baumard & Osiurak (2019), bodily experience develops in everyday life under the influence of language by thinking and talking about body parts and actions. Investigations about the relationship between language and action demonstrates the involvement of motor systems in the processing of action-related language (Crivelli et al., 2018; Dalla Volta et al., 2009; Shebani & Pulvermüller, 2018). Shebani and Pulvermüller (2018) hypothesised that processing of action words semantically related to complex actions (e.g. citing "finger" and "grasping") might facilitate elementary movements, by pre-activating a part of the movement circuit. In this context, it would also be desirable to examine if explicit conversations about body parts ("body talk") might benefit the development of BI in children with HCP.

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4.4. Selective impairment of body representation levels in children with hemiplegic cerebral palsy: bottom-up and top-down classification approaches

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Abstract

Studies on adults with focal brain damage indicated that body representation seems organized into three levels: body schema (BS), body structural description (BSD), and body image (BI). However, only little is known about potential dissociations of these levels of body representation in children. Therefore, this study aimed at investigating whether children with hemiplegic cerebral palsy (HCP) present selective impairments at specific levels of body representation. We used a combination of data- and theory-driven analyses an data of 73 children with hemiplegic cerebral palsy (mean age=9.03 [sd=2.48] years) and 141 typically developing children (mean age=8.17 [sd=1.82] years). Multivariate cluster analyses indicated four subgroups with different profiles across levels of body representation: i) a cluster with specifically high scores on BI, ii) a cluster presenting consistently low scores on BS, BSD, and BI, iii) a cluster with selective impairment in BS, and iv) a cluster of children with spared body representation. Using methods to evaluate single cases, we identified 22 cases of selective impairments across all three body representation levels in children with HCP which substantiated results of the cluster analysis. Moreover, the pattern of dissociations we observed here is consistent with results observed previously in adults. To the best of our knowledge, this is the first study employing a combined data-driven bottom-up and theory-driven top-down approach to identify profiles of body representation of selective impairments of body representation in children with HCP.

Keywords: body representation; body schema; body structural description; body image; hemiplegic cerebral palsy; top-down approach; bottom-up approach; cluster analysis.

Introduction

Experiences from somatosensory, proprioceptive, visual, auditory, olfactory, vestibular, visceral and motor-related systems are essential for the development of mental representations of one's own body and, as a consequence also for action execution, self-representation and social interactions (Berlucchi & Aglioti, 2010; de Vignemont, 2010; Head & Holmes, 1911; Longo & Haggard, 2012[a,b]).

Based on studies on adults with focal brain damage (Coslett, 1998; Schwoebel & Coslett, 2005; Sirigu et al., 1991) and further supported by developmental studies (Camões-Costa et al., 2010; Slaughter & Brownell, 2012), the neuropsychological literature indicates that body representation is organized into three interactive but partially segregable levels: i) body schema (BS) related to the online and dynamic proprioceptive representation of body posture and movement; ii) body structural description (BSD) related to the visual representation of the body and relations between its parts; and iii) body image (BI) related the lexical-semantic and affective meanings of the body and its parts (cf. Coslett, 1998; Sirigu et al., 1991).

Specific patterns of impairment at each of these levels are well documented by behavioral and neuroimaging studies as well as neuropsychological case studies in adults who suffered brain lesions (Buxbaum & Coslett, 2001; Frassinetti et al., 2008; Frassinetti et al., 2010; Schwoebel & Coslett, 2005; Sirigu et al., 1991). Buxbaum and Coslett (2001) presented a patient with an autotopagnosic patient with left hemispheric brain damage showing selective impairments in pointing to body parts on himself or others and thus in BSD. However, this patient performed well when asked to point to parts of animals and inanimate objects. Moreover, the patient also performed well in tasks assessing BS and BI (Buxbaum & Coslett, 2001). Sirigu et al. (1991) also reported a selective impairment of BSD, in an autotopagnosic patient with diffuse cerebral atrophy. Despite do not assessed BS, Sirigu et al. (1991) assessed BI level and the autotopagnosic patient performed well in tasks assessing BI. Examining more than 70 stroke patients, Schwoebel & Coslett (2005) found 18 patients with selective deficits of body representation. In particular, two patients showed selective deficits in BI, three patients with selective deficits in BSD, and 13 patients presented with selective deficit in BS.

It is worth noting, however, that despite the wealth of studies devoted to understand the selectivity of impairments observed for body representation in adults, to date only little is known about the potential dissociations of different levels of body representation in children. Although not as frequent as for adults, patterns of selective body representation impairments have been described in children with brain lesions (Frassinetti et al., 2012; Guedin et al., 2018; Nuara et al., 2019), medullary lesions (Salvato et al., 2017) and with developmental coordination disorder (Adams et al., 2017; Fuelscher et al., 2015, Fuelscher et al., 2016).

Importantly, however, it was argued that dissociations among specific neuropsychological impairments in children may be masked by functional and structural reorganization of the developing brain (Krägeloh-Mann et al., 2017; Staudt, 2010[a]; Staudt, 2010[b]). The study conducted by Fontes et al. (2017) was the first to investigate the three levels of body representation in HCP comparing their performance to the performance of typically developing children (TD). In this context, it is important to note that children with hemiplegic cerebral palsy (HCP) were found to present impairments at all three different levels of body representation (Fontes et al., 2014; Fontes et al., 2017).

As such, it is not clear yet, whether difficulties presented by children with HCP regarding body representation may i) reflect more general effects of damage and remission of the developing brain (as suggested by Fontes et al., 2017) or ii) indeed indicate specific patterns of impairments of body representation. In line with this evidence, and considering the high heterogeneity of deficits among children with HCP, it seems possible that children with HCP present patterns of impairments of body representation that may be selective to specific levels of body representation as described above for adults.

Accordingly, this study aimed at investigating whether children with HCP present selective impairments at specific levels of body representation. Therefore, performance of 73 children with HCP in tasks assessing body representation was compared to that of 141 typically developing (henceforth TD) children. Two strategies were employed to identify possible selective impairments of body representation: multivariate classification at the group level and evaluation at the single-case level.

Methods

Participants

Participants were 214 children, aged from 5 to 16 years with typical development or HCP. The group of TD children comprised 141 children aged 5-13 years (mean age=8.17 [sd=1.82] years). The group with HCP comprised 73 children aged 5-16 years (mean age=9.03 [sd=2.48] years). In the group of children with HCP, 41 had right HCP with an age range of 5-16 years (mean age=9.15 [sd=2.36] years), and 38 had left HCP with an age range of 5-16 years (mean age=8.88 [sd=2.64] years). Participants were recruited from similar socioeconomic backgrounds in free-of-charge stated-runned schools and rehabilitation centers in the state of Minas Gerais, Brazil. All children scored above the 15th percentile on a test of general cognitive ability and completed a battery of neuropsychological assessments. Data from all children were considered for the analysis. The study was approved by the local research ethics board (COEP–UFMG) in compliance with the Helsinki principles. Informed consent was obtained in written form from parents prior to the study and oral assent from children prior to testing.

Instruments

Neuropsychological assessments included general cognitive ability, motor dexterity, and tasks evaluating the three levels of body representation (i.e., BS, BI, and BSD). All tasks are described in the following.

General cognitive abilities: were assessed using the Raven's Coloured Progressive Matrices (CPM – Angelini et al., 1999) validated for the Brazilian population (up to 12 years) and the subscales Vocabulary and Block Designs of the Wechsler Intelligence Scale for Children (WISC-IV - Rueda et al., 2013) for children over 12 years of age. The analyses were based on z-scores (M = 0, SD = 1), calculated as described in the respective manual.

Motor Dexterity: Motor dexterity was examined using the Nine Hole Peg Test (9-HPT; Mathiowetz et al., 1985; Poole et al., 2005). The pegboard was centered in front of the participant with the container side on the same side as the hand being tested. Participants were instructed to pick up the pegs (one at a time), put them into the holes and then, remove the pegs one at a time and return them to the container as faster as they could. The dominant hand was tested first and non-dominant hand was tested next. Each run was timed and task solution times were recorded.

Assessment of Body Representation: Neuropsychological assessment of body representation was based on a triadic model, comprising tasks at the level of BS, BSD and BI (Coslett, 1998; Sirigu et al., 1991). Tasks employed in the current study were used in prior studies to assess the respective levels of body representation (cf. Fontes et al., 2014; Fontes et al., 2017; Salvato et al., 2017, Souto et al., 2020[a]; Souto et al., 2020[b]), and are described below.

Assessment of Body Schema (BS)

There were three tasks used to assess body schema:

i) **Hand Laterality** (cf. Fontes et al., 2014). Drawings of 12 single hands were presented on a computer screen. The child was instructed to indicate whether each picture was of a right hand or a left hand (there were six right and six left hands displayed). The hand-stimuli appeared in different positions (dorsal, palmar, and laterally rotated views of a hand with the fingers pointing medially or downward). Responses were awarded one point when the child correctly identified the hand (one point awarded for each hand correctly classified as right or left). Accuracy was categorized with 0 for incorrect responses and 1 for correct responses. A total score was calculated. Internal consistency estimates for this task was KR20=0.70 as reported by Fontes et al. (2014).

ii) **Imitation of Meaningful Gestures** (cf. Fontes et al., 2014). Ten digital animations of meaningful gestures were presented on the computer screen. Meaningful gestures comprised animations of "waving goodbye", "asking for silence", "military salute", "pointing straight ahead", the "OK" sign, the "no" sign, "blowing a kiss", the "stop" sign, "clapping hands", and "pointing to one side". The child was instructed to imitate the gesture, independent of hand laterality. Responses were coded as correct when the imitation correspondent to a prespecified model. Accuracy was categorized with 0 for incorrect responses and 1 for correct responses. A total score was calculated. Internal consistency estimates for this task was KR20=0.60 as reported by Fontes et al. (2014).

iii) **Imitation of Meaningless Gestures** (cf. Fontes et al., 2014). Ten drawings of meaningless gestures were presented on a computer screen. Five drawings depicted fingers in specific positions, and five pictures depicted arbitrary positions of the upper limb in relation to the trunk and head. The child was instructed to imitate these gestures, independent of hand laterality. Responses were considered correct when the imitation correspondent to the model. Accuracy was categorized with 0 for incorrect responses and 1 for correct responses. A total score was calculated. Internal consistency estimates for this task was KR20=0.66 as reported by Fontes et al. (2014).

Assessment of Body Structural Description (BSD)

Two tasks were used to assess body structural description:

i) **Matching Body Parts by Location** (Fontes et al., 2014). Stimuli consisted of digital drawings of body parts (i.e., leg, trunk, ear, foot, wrist, elbow, hand, nose, eye, hair, arm), which were presented in 11 trials on a computer screen. Stimuli in each trial consisted of four body part pictures organized in two rows, one above and three others. Children were asked to select among the three pictures in the lower row the physical continuation of the body part depicted on the above picture. For example, when the sample picture represented a leg, the response was correct when children selected the picture of a foot (among distractor images of an ear and a hand). Responses were coded as correct when children identified (i.e., pointed to) the correct body part. Accuracy was categorized with 0 for incorrect responses and 1 for correct responses. A total score was calculated. Internal consistency estimates for this task was KR20=0.60 as reported by Fontes et al. (2014).

ii) **Finger Gnosia Task** (Benton et al., 1994; Dellatolas et al., 1998). Testing was done in three steps. First, in full view of the hand, children were asked to locate single fingers touched by the examiner with the pointed end of a pencil. Second, with the hand hidden from view, children were again asked to locate single fingers touched by the examiner. Third, with the hand hidden from view, children had to locate pairs of fingers simultaneously touched by the examiner. Children should respond verbally calling out their names assisted by a drawing of a hand with fingers names (indicated by numbers). This drawing was available to them for all three steps. A total score was calculated for each child based on correct identification of fingers. Internal consistency of Finger Gnosia Task was KR20=0.79 in the original study (Dellatolas et al., 1998).

Assessment of Body Image (BI)

Two tasks were used to assess body structural description:

i) **Body Parts and Object Association** (Fontes et al., 2014). Stimuli for this task consisted of digital drawings of body parts (i.e., leg, trunk, ear, foot, wrist, elbow, hand, nose, eye, hair, arm) and objects (e.g. grooming tools or items of clothing or accessory, e.g., cap, belt, wristwatch, gloves, sweater, jeans, socks, glasses, toothbrush, hat, sneakers). Stimuli were presented in 11 trials. Each trial consisted of four pictures in two rows, with an object presented above three body parts. Children were asked to select among the three body pictures the one functionally matching the object. For example, in one trial the object was a watch. As such, the correct response was the wrist (with ankle and elbow presented as distractors). Children were instructed verbally to “point to or to say the name of the body part that is related to the object presented”. Responses were coded as correct when the child identified (pointed to or verbally indicated) the matching body part correctly. Accuracy was categorized with 0 for incorrect responses and 1 for correct responses. A total score was calculated. Internal consistency estimates for this task was KR20=0.60 as reported by Fontes et al. (2014).

ii) **Naming Body Parts** (Fontes et al., 2014). Stimuli consisted of digital drawings of 18 body parts (i.e., hair, belly, foot, mouth, arm, leg, knee, neck, shoulder, face, nose, head, elbow, ear, chest, eye, hand, and back) presented on a computer screen. Children were instructed verbally to “say the name of the presented body part”. Responses were coded as correct when the child correctly named the depicted body part. Accuracy was categorized with 0 for incorrect responses and 1 for correct responses. A total score was calculated. Internal consistency estimates for this task was KR20=0.63 as reported by Fontes et al. (2014).

For each trial in each task, accuracy was summed. After summing up points awarded in each task, sum scores were calculated for the BS (summing up over all 3 tasks), BSD summing up over both tasks) and BI (summing up over both tasks) measures. The sum scores of body representation measures (i.e., for BS, BSD, BI) and the speed of manual dexterity were then transformed into z-scores ($M = 0$, $SD = 1$) based on the age. The z-scores for children with HCP aged 13 years or more were calculated based on the equivalent scores of the 13-year-olders in the TD group.

Procedures

Children were assessed individually in their schools or rehabilitation centers in two sessions of approximately 60 minutes each, by specifically trained undergraduate psychology and physiotherapy students. The order of the neuropsychological tests was pseudo-randomized in two different sequences.

Statistical analyses

Statistical analyses were conducted in four steps. First, descriptive statistics compared children with left or right HCP with children with TD on sex, age, and general cognitive abilities. Second, children with HCP were individually classified using cluster analysis according to their performance on the body representation tasks. The Ward method with squared Euclidean distance was used. z-scores from the summed scores for each level of body representation (i.e., BS, BSD, BI) were used as the criterion variables for cluster formation. Third, validity of the clusters obtained was evaluated using: a) repeated-measures ANOVA to appraise the hypothesis that clusters reflect differences in levels of body representation; b) ANCOVA controlling for general cognitive ability to compare performance of children in each cluster with TD children, c) mixed-model analysis of variance to evaluate potential performance differences between paretic/non-dominant with the non-affected/dominant hand in the motor dexterity task and between groups (control group and clusters). Fourth, identification of children with HCP presenting specific impairments in a body level, comparing their performance to that of TD children (Crawford & Garthwaite, 2002; Crawford & Howell, 1998; Crawford et al., 2010).

Results

Results will be presented in four sections: a) sociodemographic characteristics; b) classification of body representation impairment patterns c) validity of body representation impairment patterns; d) selective impairments in body representation.

Sociodemographic characteristics

In general, children with HCP were significantly older ($t_{[212]}$=-2.87, p<0.01, d=0.40) and had significantly lower general cognitive abilities ($t_{[212]}$=6.22, p<0.01, d=0.93) than children with TD. No group differences were observed for gender. Moreover, analyses indicated that children with left HCP and right HCP did not differ significantly in their scores on general cognitive ability as well as the semantic word fluency. Therefore, these two groups were pooled for further analyses. Descriptives for the children with TD and HCP are presented in Table 1.

Table 1. Characteristics of the sample.

	TD (n=141)			RHCP (n=39)			LHCP (n=34)			ANOVA							
	n (%)	**m**	**sd**	**n (%)**	**m**	**sd**	**n (%)**	**m**	**sd**	**F (2;211)**	**p**	**partial η^2**	**Post-hoc (Bonferroni)**	**d**	**χ^2**	**p**	**ϕ**
Female (Gender)	66 (46.8)	-		19 (48.7)	-		18 (52.9)	-		-	-	-	-	-	0.811	0.59	0.44
Age	141 (100)	8.17	1.82	39 (100)	9.15	2.36	34 (100)	8.88	2.64	4.259	<0.01	0.039	TD<RHCP TD=LHCP RHCP=LHCP	0.47 0.31 0.11	-	-	-
Intelligence (z- score)	141 (100)	0.43	0.77	39 (100)	-0.26	0.69	34 (100)	-0.18	0.57	19.395	<0.001	0.155	TD>RHCP TD>LHCP RHCP=LHCP	0.94 0.90 0	-	-	-

TD = typically developing children group; RHCP = right hemiplegic cerebral palsy; LHCP = left hemiplegic cerebral palsy.

Classification of body representation impairment patterns

To identify possible subgroups of body representation performance that may eventually be associated with selective impairments, we used a bottom-up strategy employing a cluster analysis to identify candidate subgroups and evaluating their validity afterwards.

As criteria for cluster membership, we used performance scores for BS, BSD and BI. Four clusters were identified using dissimilarity coefficients obtained at each stage of the agglomeration processes as criteria to select the final solution for the analysis. Figure 1 exhibits the dendogram obtained during the assignment of individuals to clusters.

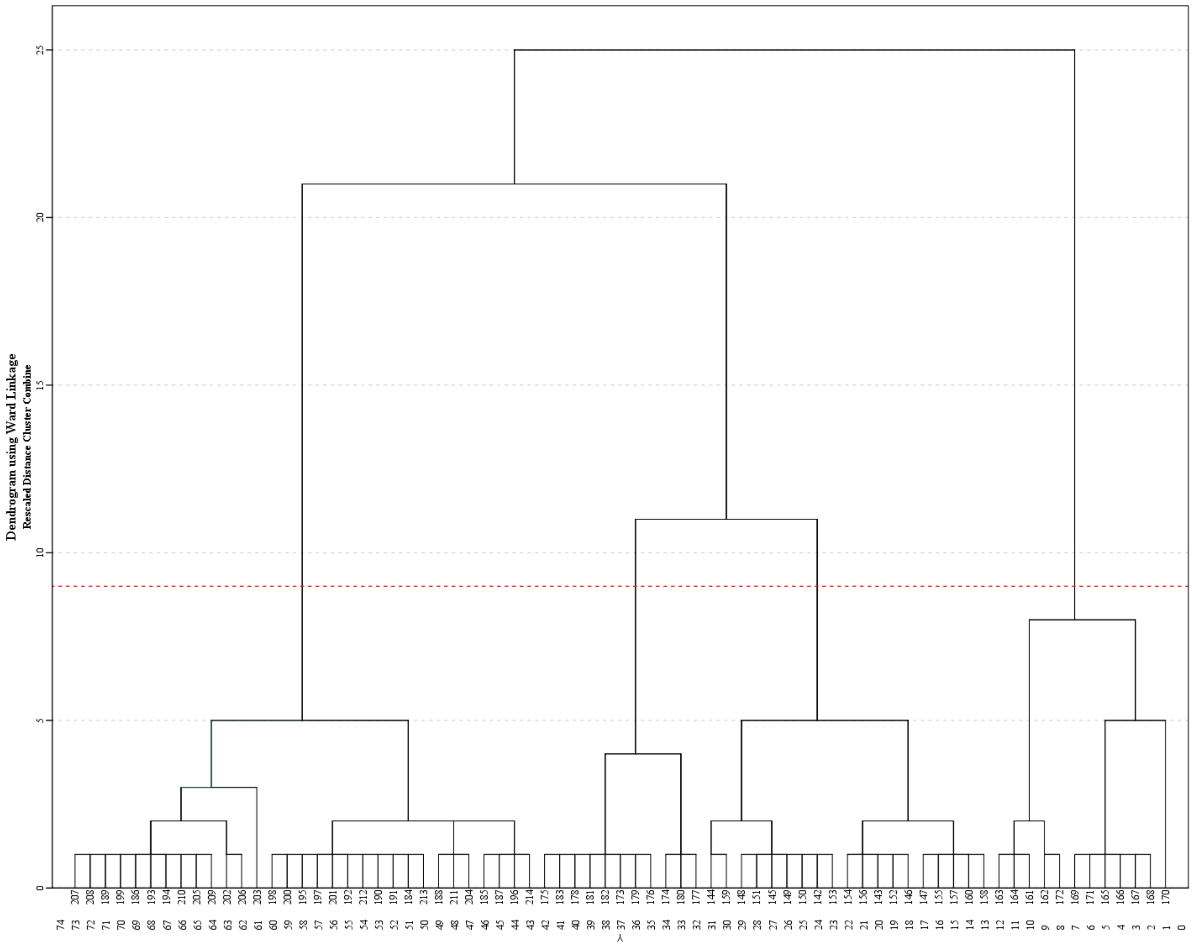

Figure 1. Dendogram showing the formation of clusters. Red line indicates the cut off for the clusters considered (i.e. four).

Cluster 1 was formed by 19 children (12 with RHCP and 7 with LHCP; mean age=8.58 [sd=2.67] years), Cluster 2 was formed by 12 children (5 with RHCP and 7 with LHCP; mean age=8.17 [sd=1.85] years), Cluster 3 was formed by 11 children (5 with RHCP and 6 with LHCP; mean age=8.18 [sd=1.66] years), and Cluster 4 was formed by 31 children (17 with RHCP and 14 with LHCP; mean age=9.94 [sd=2.62] years). Importantly, the clusters did not differ significantly with regard to the distribution of gender ($\chi^2_{[3]}$=2.223, p=0.53, ϕ=0.175), general cognitive abilities ($F_{[3;69]}$=0.819, p=0.49, η^2_p=0.034), age ($F_{[3;69]}$=2.654, p=0.06, η^2_p=0.103) and laterality of paresis ($\chi^2_{[3]}$=1.696, p=0.64, ϕ=0.152).

Each cluster was interpreted and characterized according to performance on the respective criterion variables (i.e., scores on BS, BI, and BSD). Figure 2 illustrates performance profiles of children from each cluster on the criterion variables. Descriptive for each cluster with means and standard deviations for each criterion variable are given in Table 2. To characterize the clusters, rANOVAs (with BS, BSD, and BI as factor) were calculated discerning performance on all three body representation levels followed by Bonferroni-corrected pairwise comparisons.

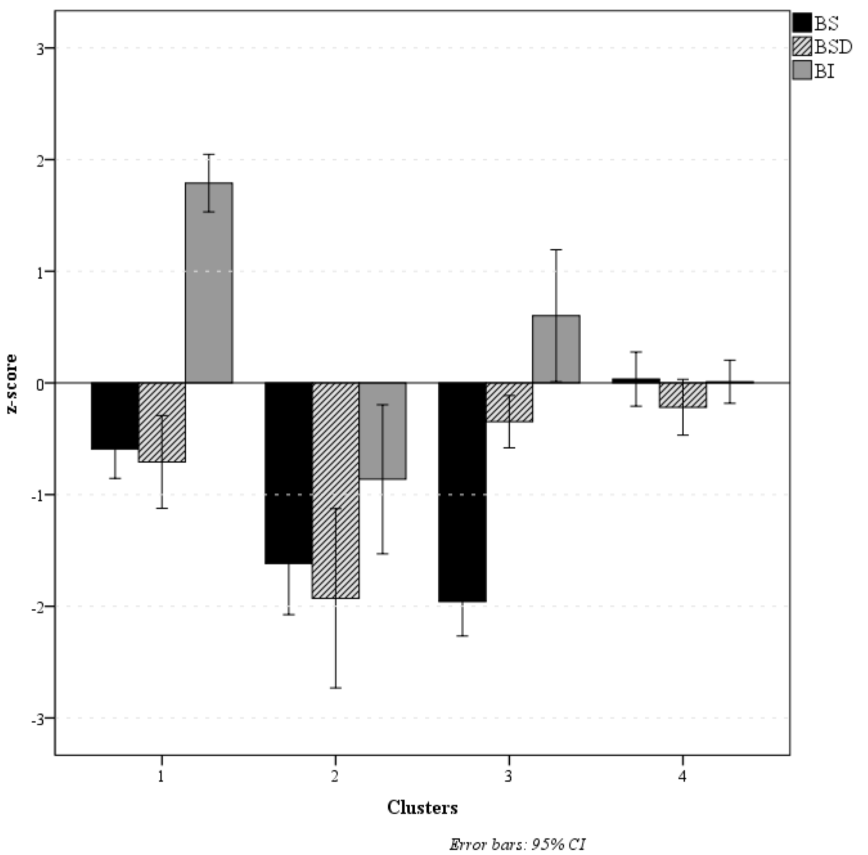

Figure 2. Performance on criterion variables across clusters. *Note:* Cluster 1 reflected by very high performance on BI, and low performance on BS and BSD (henceforth high BI cluster). Cluster 2 indicates low to very low performance in all domains of body representation (henceforth general impairment cluster). Cluster 3 was associated with very low performance on BS (henceforth selective BS impairment cluster), whereas performance for participants in Cluster 4 was average for all domains of body representation (henceforth spared body representation cluster).

Table 2. Repeated measures analysis of variance (rANOVA) among body representation domains within clusters

	BS	BSD	BI	rANOVA			Post-hoc (Bonferroni)
	mean (sd)	mean (sd)	mean (sd)	F	P	partial η^2	
Cluster 1	-0.59 (0.54)	-0.70 (0.86)	1.78 (0.53)	117.548	<0.001	0.867	BI > BS = BSD
Cluster 2	-1.61 (0.72)	-1.92 (1.26)	-0.86 (1.05)	3.283	<0.078	0.230	-
Cluster 3	-1.95 (0.45)	-0.34 (0.34)	0.60 (0.87)	41.693	<0.001	0.807	BI > BSD > BS
Cluster 4	0.03 (0.66)	-0.21 (0.68)	0.01 (0.52)	1.651	<0.202	0.052	-

BS = body schema; BSD = body structural description; BI = body image. Cluster 1 = high BI cluster; Cluster 2 = general impairment cluster; Cluster 3 = selective BS impairment cluster; Cluster 4 = spared body representation cluster.

Table 3. Descriptive data and analysis of variance (ANOVA) and covariance (ANCOVA) among control group and clusters

	Control group	Cluster 1	Cluster 2	Cluster 3	Cluster 4	ANOVA			Post-hoc (Bonferroni)
	mean (sd)	mean (sd)	mean (sd)	mean (sd)	mean (sd)	F (4,209)	p	η^2	
Age	8.17 (0.17)	8.57 (0.46)	8.16 (0.58)	8.18 (0.61)	9.93 (0.36)	4.929	<0.001	0.086	Control group = Cluster 1 = Cluster 2 = Cluster 3; Cluster 4 > Control group; Cluster 1 = Cluster 2 = Cluster 3 = Cluster 4.
Intelligence	0.43 (0.77)	-0.09 (0.60)	-0.27 (0.49)	-0.46 (0.69)	-0.20 (0.68)	10.097	<0.001	0.162	Control group > Cluster 1 = Cluster 2 = Cluster 3 = Cluster 4
	Control group	**Cluster 1**	**Cluster 2**	**Cluster 3**	**Cluster 4**	**ANCOVA**			**Post-hoc (Bonferroni)**
	mean (sd)	**mean (sd)**	**mean (sd)**	**mean (sd)**	**mean (sd)**	**F (4,207)**	**p**	**η^2**	
BS	0.36 (0.75)	-0.59 (0.54)	-1.61 (0.72)	-1.95 (0.45)	0.03 (0.66)	40.843	<0.001	0.441	Control group = Cluster4 > Cluster 1 > Cluster 2 = Cluster 3
BSD	0.33 (0.78)	-0.70 (0.86)	-1.92 (1.26)	-0.34 (0.34)	-0.21 (0.68)	21.911	<0.001	0.297	Control group = Cluster 3 = Cluster 4 > Cluster 1 > Cluster 2
BI	-0.21 (0.80)	1.78 (0.53)	-0.86 (1.05)	0.60 (0.87)	0.01 (0.52)	32.002	<0.001	0.382	Cluster 1 > Cluster 3 > Control group = Cluster 4 > Cluster 2

BS = body schema; BSD = body structural description; BI = body image. Cluster 1 = high BI cluster; Cluster 2 = general impairment cluster; Cluster 3 = selective BS impairment cluster; Cluster 4 = spared body representation cluster.

Cluster 1 - High BI cluster: Performance in BI, was significantly higher than that in BS (p<0.001; d=4.43) and BSD (p<0.001; d=3.47). No differences in performance were observed between BS and BSD (p=1.00; d=0.15).

Cluster 2 – general impairment cluster: Performance in all body representation tasks (BS, BSD and BI) was below average (all z-scores < .08), with no significant differences between levels of body representation.

Cluster 3 – selective BS impairment cluster: Performance in BS was significantly lower as compared to performance in BSD (p<0.001; d=4.04) and BI (p<0.001; d=3.68). Performance in BSD was significantly lower than performance in BI (p<0.01; d=1.42).

Cluster 4 – spared body representation cluster: Performance in all three-body representation levels was average, with no significant differences between performance on levels of body representation.

Validity of body representation impairment patterns

We ran ANOVAs to evaluate difference in age and general cognitive ability between the control group and children of the respective clusters (Table 3). Children from the *spared body representation cluster* were significantly older from the control group with regard to age (p<0.001; d=6.25). With respect to general cognitive abilities, the control group showed significantly higher general cognitive abilities than children in the *high BI cluster* (p<0.03; d=0.75), the *general impairment cluster* (p<0.01; d=1.08), the *selective BS impairment cluster* (p<0.001; d=1.22), and the *spared body representation cluster* (p<0.001; d=0.87). There were no other significant difference for age or general cognitive abilities between clusters and the control group (all p>0.05)

To evaluate the validity of the clusters, we analyzed differences between children in the respective clusters and the control group regarding all three levels of body representation. Due to above reported significant differences in age and general cognitive ability we ran ANCOVAs considering general cognitive abilities and age as covariates. Table 3 gives descriptives with means, standard deviations and the ANCOVA results for the comparison of each cluster with the control group.

Body schema

Children of the *spared body representation cluster* did not differ from those of the control group (p=0.57, d=0.47), but scored higher than children from the *high BI cluster* (p<0.02, d=1.04), the *general impairment cluster* (p<0.001, d=2.37), and the *selective BS impairment cluster* (p=0.001, d=3.51). Children of *high BI cluster* showed better performance than children of the *general impairment cluster* (p<0.001, d=1.60) and the *selective BS impairment cluster* (p<0.001, d=2.74). Performance of children in the *general impairment cluster* did not differ significantly from that of children in the *selective BS impairment cluster* (p<0.99; d=0.57). Children of the control group performed better than children of the *high BI cluster* (p<0.001; d=1.45), the *general impairment cluster* (p<0.001; d=2.68), and the *selective BS impairment cluster* (p<0.001; d=3.74).

Body structural description

Children in the *general impairment cluster* performed more poorly than children of the control group (p<0.001, d=2.15), the *high BI cluster* (p<0.001, d=1.13), the *selective BS impairment cluster* (p<0.001, d=1.71), and the *spared body representation cluster* (p<0.001, d=1.69). In contrast, there were no significant differences between the control group and children from the *selective BS impairment cluster* as well as the *spared body representation cluster* (both p>0.05). Children of the *high BI cluster* significantly performed lower than those in the control group (p<0.001, d=1.25).

Body image

Children in the *high BI cluster* performed better than those of the control group (p<0.001, d=2.93), the *general impairment cluster* (p<0.001, d=3.17), the *selective BS impairment cluster* (p<0.001, d=1.64), and the *spared body representation cluster* (p<0.001, d=3.37). Moreover, children in the *selective BS impairment cluster* scored higher than the control group (p<0.03, d=0.97) and children in the *general impairment cluster* (p<0.001, d=1.51), but did not differ from those in the *spared body representation cluster* (p=0.36, d=0.82). Children of the *general impairment cluster* performed worse than those in the *spared body representation cluster* (p<0.01, d=1.05) as well as in the control group (p<0.03, d=0.70). The difference between the control group and children in the *spared body representation cluster* was not statistically significant ($p<.99$, d=0.33).

Differences in motor dexterity regarding different patterns of body representation impairments

We also evaluated potential differences in motor dexterity between affected and non-affected hands according to the different patterns found in our clusters (Figure 3). The mixed-model ANCOVA with general cognitive ability and age as covariates discerned the influences of the independent between-participants variable participant group (i.e., control group *vs. high BI cluster vs. general impairment cluster vs. selective BS impairment cluster vs. spared body representation cluster*) and the within-participant variable motor dexterity (i.e., dominant/non-affected hand *vs.* non-dominant/affected hand).

Results indicated a significant main effect of participant group ($F_{[4;188]}$=57.849; $p<0.001$, η^2_p= 0.552). Bonferroni-corrected pairwise comparisons indicated that scores of children with HCP in all clusters differed from the control group (all $p<0.001$). Additionally, children in *spared body representation cluster* performed significantly better than children in the *high BI cluster*, the *general impairment cluster*, and the *selective BS impairment cluster* (all $p<0.05$), whereas there were no significant differences between children in the *high BI cluster*, the *general impairment cluster* and the *selective BS impairment cluster*.

There also was a significant main effect of manual dexterity ($F_{[1;188]}$=69.966; $p<0.001$, η^2_p = 0.271). Pairwise comparisons indicated that motor dexterity performance was better for the dominant/non-affected hand than for non-dominant/affected hand ($p<0.001$). Influences of the covariates were not significant (age p=0.058, general cognitive ability p=0.20).

Additionally, the interaction of participant group and manual dexterity was significant ($F_{[4;188]}$=48.165; $p<0.001$, η^2_p=0.506). To evaluate where this interaction originated from, we followed the procedure suggested by Kirk (2013). First, we computed, for each participant, the effect of the motor dexterity as the difference between their performance for dominant/non-affected hand as well as for non-dominant/affected hand. Then, we evaluated the influence of participant group on the respective effects of the motor dexterity by running a univariate ANCOVA (considering general cognitive ability and age as covariates). This allowed us to evaluate the influence of participant group (i.e., control group *vs. high BI cluster vs. general* *impairment cluster vs. selective BS impairment cluster vs. spared body representation cluster*). ANCOVA results indicated that the performance difference between the dominant/non-affected hand and the non-dominant/affected hand differed significantly across participant groups ($F_{[4;207]}$=18.227; p<0.001, η^2_p=0.260). Bonferroni-corrected pairwise comparisons indicated that the control group presented significantly smaller difference between the dominant/non-affected hand and the non-dominant/affected statistically than all clusters (all p<0.05). Additionally, children with HCP in the *spared body representation cluster* presented significantly smaller difference between the non-affected hand and the affected hand than children with HCP in the *high BI cluster*, the *general impairment cluster*, and the *selective BS impairment cluster* (all p<0.05). However, there were no significant differences between children of the *high BI cluster*, the *general impairment cluster*, and the *selective BS impairment cluster* (all p>.99).

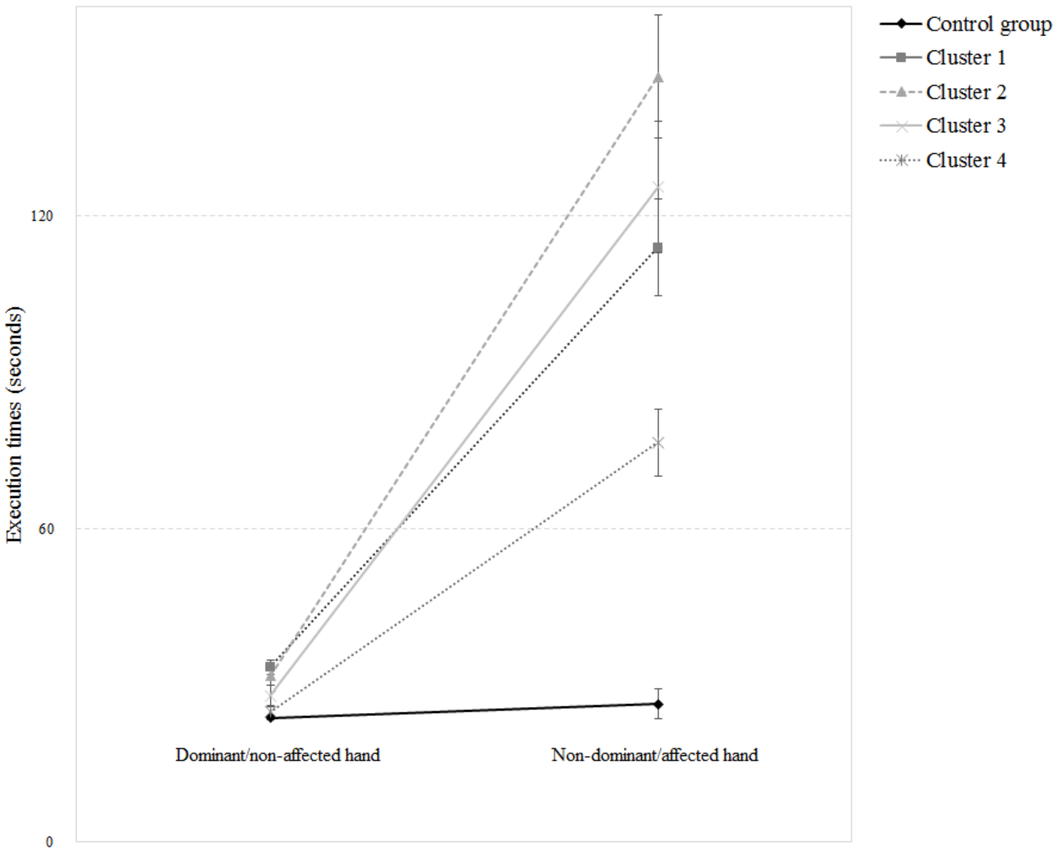

Figure 3. Interaction diagram demonstrate group (control group and clusters) differences in motor dexterity. Cluster 1 = high BI cluster; Cluster 2 = general impairment cluster; Cluster 3 = selective BS impairment cluster; Cluster 4 = spared body representation cluster. Error bars indicate standard error.

Selective impairments in body representation

Cluster analysis revealed that there were different patterns of body representation deficits in children with HCP. To further investigate whether there are cases of unique selective deficits in body representation, individual performance of each child with HCP was compared with the mean performance presented by the control group, using the procedure suggested by Crawford (Crawford and Howell, 1998; Crawford and Garthwaite, 2002; Crawford et al., 2010). The analyses revealed selective impairment in body representation in 22 children with HCP (54.5% female; right HCP = 8 cases, left HCP = 14 cases). Five children with selective impairments were from the *high BI cluster*. Of these, 1 child had a selective impairment in BS and 4 children had a selective impairment in BSD. In the *general impairment cluster* 3 cases of selective impairment of body representation were found, 1 case for each level of body representation. Moreover, nine children with selective impairment in BS were observed in the *selective BS impairment cluster*. In the *spared body representation cluster* there were 5 cases of selective impairment, 1 for in BS and 4 in BSD. Table 4 shows descriptive data from these 22 cases of selective impairment.

Table 4. Modified t-test comparisons for individual scores for children with HCP (indicated by arbitrary participant codes).

Participant	Gender	Age (years)	Laterality of hemiparesis	Cluster	Modified t-test*											
					BS				BSD				BI			
					Score	t	p	d	Score	t	p	d	Score	t	p	d
#125	M	5	L	1	-1.10	1.94	**0.02**	-1.94	-0.32	0.83	0.20	-0.83	0.79	1.25	0.10	1.25
#063	M	5	L	1	-0.12	0.64	0.26	-0.64	-1.29	2.06	**0.02**	-2.07	2.33	3.15	*<0.01*	3.16
#079	M	10	L	1	-0.49	1.12	0.13	-1.12	-1.18	1.93	**0.02**	-1.94	2.35	3.18	*<0.01*	3.19
#090	F	7	R	1	-0.56	1.22	0.11	-1.23	-1.42	2.23	**0.01**	-2.24	2.18	2.97	*<0.01*	2.98
#134	F	6	R	1	-0.69	1.39	0.08	-1.39	-1.36	2.15	**0.01**	-2.16	1.66	2.32	*0.01*	2.33
#207	M	9	L	2	-1.62	2.61	**<0.01**	-2.62	-0.28	0.78	0.21	-0.79	-0.94	-0.90	0.18	-0.90
#148	F	10	R	2	-0.43	1.05	0.14	0.29	-1.26	2.02	**0.02**	-2.03	-0.75	-0.65	0.25	-0.66
#084	M	7	R	2	-0.64	1.32	0.09	-1.32	-0.80	1.44	0.07	-1.45	-1.83	-1.99	**0.02**	-2.00
#001	M	8	L	3	-2.13	2.59	**<0.01**	-2.60	-0.29	0.79	0.21	-0.79	0.06	0.35	0.36	0.35
#009	F	9	R	3	-3.13	4.61	**<0.01**	-4.63	-0.42	0.96	0.16	-0.96	2.45	3.30	*<0.01*	3.31
#118	F	8	L	3	-1.86	2.94	**<0.01**	-2.95	-0.32	0.83	0.20	-0.83	-0.14	0.08	0.46	0.08
#159	F	8	L	3	-1.86	3.28	**<0.01**	-3.30	-0.35	0.87	0.19	-0.87	1.77	2.45	*<0.01*	2.46
#177	F	8	R	3	-1.86	2.94	**<0.01**	-2.95	-0.33	0.85	0.19	-0.85	0.65	1.07	0.14	1.08
#179	M	11	L	3	-1.80	2.86	**<0.01**	-2.87	-0.22	0.71	0.23	-0.71	-0.07	0.17	0.43	0.17
#182	F	10	L	3	-1.91	2.99	**<0.01**	-3.00	0.31	0.02	0.48	-0.02	1.28	1.86	*0.03*	1.86
#183	F	8	R	3	-1.86	2.94	**<0.01**	-2.95	-0.30	0.81	0.20	-0.81	0.60	1.01	0.15	1.02
#198	M	6	L	3	-1.82	2.89	**<0.01**	-2.90	-0.33	0.84	0.20	-0.84	-0.01	0.24	0.40	0.24
#095	M	11	L	4	-0.87	1.63	**0.05**	-1.64	-0.22	0.71	0.23	-0.71	-0.33	-0.15	0.44	-0.15
#006	F	6	L	4	0.60	0.31	0.37	0.31	-1.73	2.62	**<0.01**	-2.63	0.94	1.43	0.07	1.44
#030	M	9	L	4	1.10	0.97	0.16	0.98	-1.42	2.23	**0.01**	-2.23	0.15	0.46	0.32	0.46
#144	F	10	L	4	-0.55	1.21	0.11	-1.22	-3.84	5.31	**<0.01**	-5.33	-0.40	-0.22	0.41	-0.22
#152	F	9	R	4	-0.27	0.83	0.20	-0.84	-1.07	1.79	**0.03**	-1.80	0.15	0.46	0.32	0.46

*Modified t-test values were calculated based on control group data: BS = 0.36 (sd=0.75); BSD = 0.33 (sd=0.78); BI = -0.21 (sd=0.80). *CP = cerebral palsy; BS = body schema; BSD = body structural description; BI = body image.* Cluster 1 = high BI cluster; Cluster 2 = general impairment cluster; Cluster 3 = selective BS impairment cluster; Cluster 4 = spared body representation cluster. t=modified t-test proposed by Crawford and Garthwaite (2002) calculated with singlims.exe; p = power analyses; d =effect size (Crawford et al., 2010). Note: p values in **bold** indicate impaired performance (i.e., scores below the range of scores for normal controls). p values in *italic* indicate higher performance (i.e., scores higher than the range of scores for normal controls).

Discussion

The present study investigated body representation in children with HCP and its relation to specific deficits related to the three levels of body representation using bottom-up and top-down approaches. First, using a bottom-up approach, we evaluated whether there are subgroups of children with HCP who present with different patterns of deficits in body representation using cluster analysis. Four clusters of children with specific profiles of body representation were identified and compared to a TD control group. Next, we investigated potential differences between the identified clusters for motor dexterity of both the paretic hand and non-paretic hand. Last, by applying a top-down approach, we examined whether children with HCP present impairments at a specific level of body representation and evaluated whether selective impairments were associated specifically with the clusters identified above.

This is one of the first studies to employ a bottom-up approach aiming at dissociating different profiles of the three levels of body representation in children with HCP using cluster analysis. The data-driven or bottom-up approach consists of letting the groups emerge from multivariate techniques of classification (Salvador et al., 2018). In the present study, this resulted in subgroups with different profiles reflecting with intragroup similarities and between-group differences on criterion variables BI, BS, and BSD. We found a final solution of four distinct clusters: i) the *high BI cluster* of children with specifically high scores on BI and lower performance in BS and BSD; ii) the *general impairment cluster* of children presenting with consistently low performance in BS, BSD and BI; iii) the *selective BS impairment cluster* showed selective low performance in BS, and iv) the *spared body representation cluster* of children who showed average performance in all representational levels.

All clusters included children with right and left HCP. These results of the present study indicates that different profiles of specific impaired and/or spared levels of body representation may not be due to the laterality of hemiparesis, but may be related to potential (re)organization after early brain lesions and to the heterogeneity of the clinical condition frequently presented by children with HCP. In agreement with this hypothesis, the study by Fontes et al. (2017) also found impairments in BS, BSD and BI and thus all levels of body representation in children with HCP, and did not observe specific effects of the laterality of hemiplegia on impairments of body representation. However, studies of children with HCP have not yet provided clear evidence of hemisphere dominance in relation to the impairment of body representations.

Contrasting with our results, some studies observed greater impairments in BS and in motor planning in children and adolescents with right hemiplegia (Chinier et al., 2014; Crajé et al., 2010; Mutsaarts et al., 2007). According to studies of brain injury in adults, the cortical regions related to the body representations are present in both brain hemispheres, but there is evidence of right-hemisphere dominance (Berlucchi & Aglioti, 2010; Schwarzlose et al., 2005).

It is important to note that general cognitive abilities was comparable across clusters of impaired body representation whereas the control group performed significantly better than children with HCP. This is in line with previous studies that also observed lower general cognitive abilities in children with HCP compared to controls (e.g., Ashcraft et al., 1992; Fontes et al., 2017; Levine et al., 2005; Muter et al., 1997; Stiles et al., 1997; Thevenot et al., 2014; Tillema et al., 2008; Trauner et al., 2001; Trauner, 2003). Importantly, some authors argue that general cognitive abilities in children with HCP may actually be underestimated by using tests standardized on TD children who do not have motor, communication and/or visual impairments, frequently presented by children with HCP, which may well influence test results (e.g., Ballester-Plané et al., 2016; Sherwell et al., 2014; Yin Foo et al., 2013;). However, if any item is modified to make it more appropriate for a physical impairment, the item may lose the capacity to evaluate cognitive abilities (Yin Foo et al., 2013). To minimize this recurring problem in neuropsychological studies in children with HCP, effects of general cognitive abilities were controlled in this study.

As a next step of our bottom-up approach, we aimed at validating the four profiles identified in the cluster analysis evaluating differential patterns of dissociations and associations. Therefore, we compared performance on BS, BI, and BSD tasks of children from the four clusters of children with HCP (i.e., *high BI cluster*, *general impairment cluster*, *selective BS impairment cluster*, and *spared body representation cluster*) to that of the control group composed of TD children.

We observed that children from the *high BI cluster* performed significantly better than children from all other clusters and the control group with regard to BI. Moreover, concerning BS and BSD, children from "high BI cluster" performed lower than the control group and children from the "spared body representation cluster". In line with this, previous studies investigating adult's body representation following brain lesions found dissociations between BI and BSD (Buxbaum & Coslett, 2001; Sirigu et al., 1991). Considering that we tested children a potential explanation for this pattern of results may be that language networks, related to lexical-semantic body knowledge, develops during infancy and childhood is represented bilaterally in the brain (Staud, 2010[a]; Staud, 2010[b]). Due to a crowding effect after brain lesion, language abilities seem to reorganize and be largely intact, but patients may still suffer from visual-spatial deficits (Krägeloh-Mann et al., 2017;).

Performance of children in the *general impairment cluster* was lower than for those from the *high BI cluster*, the *spared body representation cluster*, and the control group for all levels of body representation. There was an exception for the difference between performance of the *general impairment cluster* and the *selective BS impairment cluster* for BS, which was not significant. As regards BSD and BI, children from the *general impairment cluster* performed worse than those from the *selective BS impairment cluster*. Considering the importance of body representation to motor planning and motor execution, self-representation and social interactions (Berlucchi and Aglioti, 2010; de Vignemont, 2010; Mutsaarts et al., 2007), this cluster might represent a group with major global deficits.

Another cluster of children presented selective impairments in body representation. As such, children from the *selective BS impairment cluster* performed worse than the control group, children from the *spared body representation cluster* and the *high BI cluster*, but did not differ from children in the *general impairment cluster* with respect to BS. The selective impairment in BS is of clinical relevance because BS has a special relationship with bodily actions, and without accurate representation of one's own body parameters, hardly any successful actions are possible (de Vignemont, 2010). Thus, impairment of BS may result in apraxia, a clinical syndrome defined as a deficit in the control of deliberate motor actions (Goldenberg, 2013). Probably, children in this group also present motor neglect, a peculiar manifestation of unilateral neglect, in which patients fail to spontaneously use their affected limb in the absence of any motor or somatosensory deficits. In addition, BS underlies performance in motor imagery tasks (de Vignemont, 2010; Parsons, 1994). Steenbergen et al. (2009) proposed that motor imagery might be useful in training the motor neural networks after injury in the central nervous system. Souto et al. (2020[b]) observed that motor imagery training combined with physical practice was effective in improving functional performance in children and adolescents with HCP. However, it needs to be investigated whether children with selective BS impairments may perform motor imagery and whether may benefit from motor imagery training.

Finally, the *spared body representation cluster* was mostly composed of children with average performance at the three levels of body representation. Average performance in this cluster was higher than performance of children from the *selective BS impairment cluster* and the *general impairment cluster* for all levels of body representation. When compared to children from the *high BI cluster*, children in the *spared body representation cluster* performed worse only for BI. Interestingly, performance of children in the *spared body representation cluster* did not differ significantly from the performance of control group at the three levels of body representation. These results suggest that, despite the occurrence of a brain lesion, some children with HCP may not present an impairment of body representation.

Despite present different patterns of body representation, children with HCP in all clusters performed slower in the motor dexterity task and presented significantly larger differences in performance between the dominant/non-affected hand and the non-dominant/affected than children present in the control group. Some earlier studies found that children with HCP presented slower performance in motor dexterity tasks for both the affected as well as the non-affected hand compared to non-dominant and dominant hands of TD children (e.g., Fontes et al., 2017; Guedin et al., 2018).

However, the present study demonstrates that, despite performing worse than the control group in a motor dexterity task, children from the *spared body representation cluster* performed better than children from the *high BI cluster*, the *general impairment cluster*, and the *selective BS impairment cluster.* Moreover, there were no significant differences between children from the *high BI cluster*, the *general impairment cluster*, and the *selective BS impairment cluster*. This indicates differences in motor dexterity among children with HCP regarding their performances in body representation tasks. In particular, it seems as the integrity of levels of body representation (verified in *spared body representation cluster*) might attenuate difficulties in motor dexterity presented by children with HCP.

These major differences in motor dexterity between the affected and non-affected hand presented by the *high BI cluster*, the *general impairment cluster*, and the *selective BS impairment cluster*, might reflect in melokinetic (also called limb-kinetic) apraxia (Binkofski & Fink, 2005; Buxbaum & Randerath, 2018). In case kinetic memories, or the representation of movement patterns, are lost, movement trajectories are executed with reduced smoothness and precision (Buxbaum & Randerath, 2018). Also, deficits in anticipation of grip force needed to lift objects and postures needed to grasp objects were associated with damage in the temporal-parietal-occipital junction, but not with left hemisphere lesions, related to praxis actions and referred as "tool use network" (Li et al., 2011). The relationship between apraxia and anticipatory grip force scaling needs future investigations (Buxbaum & Randerath, 2018).

Last, the incidence of impairments in body representations in the present study was surprisingly high. Only about 43% of children with HCP who participated of this study were clustered in the *spared body representation cluster* and showed performance largely comparable to controls. Therefore, in our sample, about 57% of children showed any kind of impairment in body representation. Such impairments are likely to substantially disrupt everyday activities of patients; as such further exploration of body representation may have important theoretical as well as clinical implications (Schwoebel & Coslett, 2005) in general. However, for the case of HCP, body impairments of representation have received surprisingly little attention so far.

Importantly, some clinical disorders are associated with a specific patterns of dissociation (Temple, 1997). In the present study, we identified 22 cases of selective impairments in body representation in children with HCP (64% children with left HCP). In the "high BI cluster", one child had a selective impairment in BS and four children had a selective impairment in BSD. Also, in this cluster four children presented specifically high performance in BI. In the "general impairment cluster", we found one case of a specific impairment for each level of body representation. In the "selective BS impairment cluster", nine children (representing 81.8% of children in this cluster) had a selective impairment in BS. In the "spared body representation cluster", despite being a cluster with average performance on all levels of body representation in general, one child presented with a selective impairment in BS and another four children in BSD. Nevertheless, these five children represented only about 16% of children in this cluster. This demonstrates that the clusters analysis are substantiated by the individual analysis. Moreover, the pattern of dissociations found in our results is consistent with the double dissociation observed previously in adults following brain lesion (Buxbaum & Coslett, 2001; Coslett, 1998; Coslett et al., 2002; Schwoebel & Coslett, 2005; Sirigu et al., 1991).

Dissociations of impairments at different levels of body representation were previously observed in children with spinal trauma and following brain injury (Frassinetti et al., 2012; Nuara et al., 2019; Salvato et al., 2017). Comparing performance on tasks that evaluate BSD and BI indicated that 3-year-old children with spinal trauma seem to present a selective deficit in BI, but not in BSD (Salvato et al., 2017). Furthermore, brain damaged children showed a double dissociation in a task on recognizing body parts (assessing BSD). Children with right-hemispheric brain damage were found impaired processing their own but not other people's body parts, whereas children with a left-hemispheric brain damage were impaired in processing others' but not their own body parts (Frassinetti et al., 2012). Moreover, children with HCP presented upper limb asymmetries when ask draw a portrait of themselves, but did not draw such alterations when portraying other children (Nuara et al., 2019). Taken together, previous studies primarily observed dissociations between recognizing one's own *versus* body parts of other, but did not report on selective impairments in body representations in children with HCP.

A criterion for examining the applicability of the cognitive-neuropsychological model (in this study, related to impairments in body representation in children with HCP) concerns the observation of impairments in specific, dissociable forms of body representation (Temple, 1997). Taken together, the findings of the present study strongly suggest that sensorimotor (BS), structural (BSD) and lexical–semantic (BI) information about the human body seems to be represented in independent systems in children, as suggested for adults by neuropsychological studies (e.g., Schwoebel & Coslett, 2005; Sirigu et al., 1991).

Although the usefulness of dissociations for understanding developmental disorders be questioned in some studies because, during the course of development, the nature of representations may change and dissociations could disappear over time (Bishop, 1997), the dissociations found in this study are also present in cases of acquired brain lesions. Furthermore, according to Castles et al. (2014) in almost every domain of developmental disorders the dissociation cases be cases selected from the population as low performances on one skill but not in another. As such, the present study provides evidence on selective impairments in body representations levels in children with HCP, and also that, the proposed neuropsychological model for body representations based on adults processing assist us in understanding developmental body representation impairments in children.

Importantly, this research has also practical implications for the clinical rehabilitation of children with HCP. Considering the impact of body representation impairments, it would be desirable to include the evaluation of body representations in clinical practice. Future studies focusing on the interaction of impairments of body representation and functional abilities in children with HCP may contribute to a better understanding of consequences of impairments of body representations and further improve the clinical approach on children with HCP. Moreover, it is important to further investigate whether and if so how functional independence (e.g., activities of daily living and school learning) and rehabilitation may be influenced by the presence of impairments in body perception and representation.

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5. GENERAL DISCUSSION

The present dissertation aimed to investigate possible interactions among body representations during the development of BI, and to investigate distinct subtypes of impairments in body representation (especially selective deficits in body representation levels) in children with HCP. In this section, we will discuss our mains results.

The literature reviewed (Auclair & Jambaqué, 2014; Bahrick & Watson, 1985; Camões-Costa et al., 2010; Crowe & Prescott; 2003; Morita et al., 2012; Rochat & Morgan, 1995; Slaughter et al., 2002; Slaughter et al., 2012; Witt et al., 1990) allow us to formulate the hypothesis that BS influences the development of BSD and, consequently, of BI. First, in Study 1, we described the developmental structure of lexical-semantic body knowledge related to BI in children with TD. We carried out an investigation using graph theory to analyze spontaneous production of words related to body parts, and also to animals and foods. Children with TD were divided into three age groups (4-6, 7-9, and 10-12 years).

For all the semantic categories investigated, we observed differences in networks parameters among the three age groups, indicating an addition of words as the age increases. Also, our results regarding knowledge of body parts corroborate findings from previous studies demonstrating that names for the structures of the head/face and limbs are learned first, followed by the names for the joints, and, lastingly, internal organs (Auclair & Jambaqué et al., 2014; Camões-Costa et al., 2010; Crowe & Prescott, 2003; Witt et al., 1990). This indicates a trend in development consistent with the development of the BS and BSD, but the implication of sensorimotor influences on BI development was only inferred in our study, as we did not evaluated BS and BSD.

On the other hand, the results obtained did not revels a pattern of influence in development of animals and foods knowledge. According to Crowe & Prescott (2003), children tend to form groups according to their familiarity with the animals and not according classes (mammals, birds, fish, amphibians and reptiles) or habitats. We did not observed this pattern in our study.

Our findings complement previous research on development of body representation in children, which is relatively based on younger children (Bahrick & Watson, 1985; Camões-Costa et al., 2010; Morita et al., 2012; Rochat & Morgan, 1995; Slaughter et al., 2002; Slaughter et al., 2012; Witt et al., 1990). In line with previous result (Auclair & Jambaqué, 2014), our data indicate that somatosensorimotor experiences might influence lexico-semantic processing of body parts, and this influence is not limited to younger children. According to that, impairments to sensorimotor functioning may influence BI development.

Children with CP present heterogeneous condition in terms of severity of impairments. In addition to the disorders of movement and posture, children with CP often show impairments in sensation and perception (vision, hearing and other sensory modalities – Rosenbaum et al., 2007). Considering the assumption that sensorimotor functioning may influence BI development, in Study 2 we evaluated the development of BI by comparing the performance of children with typical development with that of children with HCP and investigated whether the development of BI is delayed in children with HCP.

For this purpose, we conducted a cross-sectional study involving 257 children, of which 204 were typically developed control children and 53 were children with HCP. We compared the performance on word fluency for body parts (and also for animals) of children with HCP (aged from 7 to 12 years old) to that of TD children separated into three age groups: i) 4-6 years, ii) 7-9 years, and iii) TD 10-12 years.

First of all, we analyzed the four groups performance in semantic verbal fluency task (body parts and animals categories). Compared to the control groups with same age range, children with HCP did nor presented statistically differences concerning the number of errors and word repetitions for both categories. In contrast, comparisons indicated that the number of correct words were similar for children with HCP and TD 4-6 years for both animals and body parts. Results concerning impairments in semantic word fluency tasks in children with HCP are inconclusive might be due to differences in neural reorganization, and in location and extent of neural lesions (Bottcher, 2010; Liegeois, 2004). Despite that, our results indicate that children with HCP presents a delay regarding retrieval of the semantic memory content, but not in executive functions.

To verify an influence of this possible delay in retrieval of the semantic memory content in our results regarding BI development, we also compared the production of correct words related to body parts and animals. The three groups of typically developed children produced more body parts than animals, but it does not occurred for children with HCP. This indicates that production of words related to body parts are most affected by HCP than production of words related to animals.

Second, we compared the graphical parameters of networks formed using data from body parts category. We observed positive evidence in favor of the null effect for the differences between the children with HCP and those from the TD 4-6 year group. The graphical parameters obtained for the HCP group differed significantly from the parameters obtained for the TD 7-9 years and TD 10-12 years groups.

Next, we qualitatively investigated whether BS (related sensorimotor information) and BSD (related visuospatial information) contribute to the development of BI by qualitatively comparing performance of HCP and TD 4-6 years, TD 7-9 years, and TD 10-12 years. We verified that children with HCP presented a lexical-semantic network of body parts similar to that of the youngest TD group. This is in line with and also expands previous studies which suggested that children with HCP presents lower performance in naming body parts tasks, related to BI, than TD children (Christie & Slaughter, 2009; Fontes et al., 2017).

Although joints were expected to be a part of the semantic network core of children with HCP because they were part of the semantic-lexical repertoire of children of the same age, we did not observe this in our results. Accordintg to de Vignemont et al, (2009), joints are a point of reference for the segmentation of body parts and represents an important knowledge about the structure of the human body. Importantly, the segmentation of the body into parts is considered to derive from the organization of the proprioceptive, motor and perceptual systems (de Vignemont et al., 2009). Also, the identification of joints' position (derived from proprioceptive signals) are needed to motor planning (Marini et al., 2018). Literature also points to motor planning impairments in children and adolescents with HCP (Mutsaarts et al., 2006; Souto et al., 2020).

Therefore, reduced sensorimotor and visuoperceptual experiences of body seems to contribute to a delay in the development of lexical-semantic knowledge of body parts in children with HCP. Also, our results provide evidence about the relationship between language and action, also investigated in others studies (Crivelli et al., 2018; Dalla Volta et al., 2009; Shebani & Pulvermüller, 2018).

In light of these findings, Study 1 and Study 2 can be relevant for the knowledge regarding development of body representations and provide evidence that could be relevant for the development of efficient trainings for clinical populations. Further studies should also evaluate the development of the three levels of body representation in children with HCP and better investigates possible interactions between body representations during the development.

With reference to the interactions in body representations, it does not excludes the possibility of dissociations among the three levels. Many studies investigated selectivity of impairments of body representation in adults (Buxbaum & Coslett, 2001; Frassinetti et al., 2008; Frassinetti et al., 2010; Schwoebel & Coslett, 2005; Sirigu et al., 1991). Although, little is known about dissociations of different levels of body representation in children. Some studies found selective impairments body representation in children with brain lesions (Frassinetti et al., 2012; Guedin et al., 2018; Nuara et al., 2019), medullary lesions (Salvato et al., 2017) and with developmental coordination disorder (Adams et al., 2017; Fuelscher et al., 2015, Fuelscher et al., 2016). Previous studies primarily observed dissociations between recognizing one's own *versus* body parts of other, but did not report selective impairments in body representations in children with HCP (Frassinetti et al., 2012; Nuara et al., 2019).

In Study 3, we investigated whether children with HCP present different profiles of impairments of body representation. Performance of 73 children with HCP in tasks assessing body representation was compared to that of 141 typically developing (henceforth TD) children. To identify possible subgroups of body representation performance that may eventually be associated with selective impairments, we employed a cluster analysis using performance scores for BS, BSD and BI as criteria for cluster membership. Four clusters were identified: i) the *high BI cluster* of children with specifically high scores on BI and lower performance in BS and BSD; ii) the *general impairment cluster* of children presenting with consistently low performance in BS, BSD and BI; iii) the *selective BS impairment cluster* showed selective low performance in BS, and iv) the *spared body representation cluster* of children who showed average performance in all representational levels.

First, in line with previous result (Fontes et al., 2017) our data indicated that different profiles of body representation may not be due to the laterality of hemiparesis. However, some studies contrasted with our results and observed greater impairments in BS in children and adolescents with right hemiplegia (Chinier et al., 2014; Mutsaarts et al., 2007). There is no clear evidence of hemisphere dominance in relation to the impairment of body representations in children with HCP.

Second, we observed that children with HCP could present different patterns of body representation, ranging from global impairments, selective impairments, no impairments and selective higher performance. Despite that, children with HCP in all clusters performed slower in the motor dexterity task and presented significantly larger differences in performance between the dominant/non-affected hand and the non-dominant/affected than children present in the control group. Lower performance in motor dexterity tasks for both affected and non-affected hand was also observed in previous studies (e.g., Fontes et al., 2017; Guedin et al., 2018).

However, results obtained from Study 3 demonstrates that children with an average performance in body representation tasks (belonging to *spared body representation cluster*) performed better in motor dexterity task than children with some impairment in body representation. We hypothesized, therefore, that the integrity of levels of body representation might attenuate difficulties in motor dexterity presented by children with HCP. This is in accordance to the importance of functional roles of the levels of body representation in guiding action, and especially the role of the body schema (de Vignemont, 2010).

Last, by applying a top-down approach, we identified 22 cases of selective impairments in body representation, substantiated by cluster analysis. We find selective impairments for all three levels of body representations, as well was found in adults who suffered brain lesion (Schwoebel & Coslett, 2005). This could represent an evidence of the applicability of the cognitive-neuropsychological model of body representations (based on adults processing - Sirigu et al., 1991) in understanding developmental body representation impairments in children.

We started our investigation from the hypothesis formulated by Fontes et al. (2014) that "developmental disregard" phenomenon could be partially explained by body representation impairments in children with HCP. Our findings are in line with this hypothesis, because they provide additional evidence of impairments in body representation in children with HCP. Despite that, the specific role of each level of body representation in upper-limb functionality and daily functioning in children with HCP remains unclear. Finally, future studies should also focus on functional measures (as the Assisting Hand Assessment and Jebsen–Taylor Test of Hand Function to assessment of hand function, and Canadian Occupational Performance Measure and Pediatric Evaluation of Disability Inventory to assessment of daily functioning) and it is relation to body representation performance.

In addition, it will be important to explore the benefits of constraint-induced movement therapy (CIMT) and bimanual intensive training, such as hand-arm bimanual intensive training (HABIT) protocols in relation to outcomes in body representation tasks. Studies applying those protocols revels improvements in hand function and in daily functioning in children with HCP (Brandão et al., 2012; Brandão et al., 2013; Brandão et al., 2018; Charles & Gordon, 2006; DeLuca et al., 2003; Eliasson et al., 2005). Another promising intervention for children with HCP is the use of Neurofeedback training (Ayres et al., 2004; Alves-Pinto et al., 2017). Through training with the Neurofeedback, participants learn to voluntarily regulate their electrical brain activity, which contributes to accelerate functional reorganization in the brain after injury (Alves-Pinto et al., 2017). Those protocols of intervention could also allow us to better explore the developmental causal hypotheses involved in impairments of body representation and upper-limb function in children with HCP. According to Castles et al. (2014), intervention studies represents the better way to test developmental causal hypotheses. Investigating cases who show dissociations provides an opportunity to explore hypotheses about the nature of that compromise (Castles et al., 2014).

Next steps

The research agenda on body representation impairments on children with HCP offers a wide range of possible approaches. I personally aim at continuing to explore impairments of body representation in children with HCP, but, this time exploring how body representation can affect the potential motor functions of the affected upper limb and it's relation to functional outcomes.

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6. CONCLUSION

Together, the three studies that composed this dissertation provide several contributions to the literature of body representation in hemiplegic cerebral palsy. We described the development of body image in children with typical development and children with hemiplegic cerebral palsy. In addition, we verified a delay in development of body image in children with hemiplegic cerebral palsy and that sensorimotor and visuoperceptual experiences of body seems to contribute to the development of body semantic knowledge. We also demonstrated that children with hemiplegic cerebral palsy could present different patterns of body representation and that motor dexterity seems to be influenced by the preservation of body representation. It was also shown that selective cases of body representation also occurs in children, as demonstrated in studies with adults. Accordingly, our results demonstrate the multidimensionality and specificity of body representations in children with hemiplegic cerebral palsy and emphasize the need of more research on this extremely relevant topic of body representation.

APPENDIX - Publications in scientific journals during the doctoral period

Appendix 1

Fontes, P., Cruz, T., Souto, D. O., Moura, R., & Haase, V. G. (2017). Body representation in children with hemiplegic cerebral palsy. *Child neuropsychology : a journal on normal and abnormal development in childhood and adolescence*, *23*(7), 838–863. https://doi.org/10.1080/09297049.2016.1191629

Appendix 2

Souto, D. O., Cruz, T., Fontes, P., Batista, R. C., & Haase, V. G. (2020). Motor Imagery Development in Children: Changes in Speed and Accuracy With Increasing Age. *Frontiers in pediatrics*. 8, 100. https://doi.org/10.3389/fped.2020.00100

Appendix 3

Souto, D. O., Cruz, T., Coutinho, K., Julio-Costa, A., Fontes, P., & Haase, V. G. (2020). Effect of motor imagery combined with physical practice on upper limb rehabilitation in children with hemiplegic cerebral palsy. *NeuroRehabilitation*, *46*(1), 53–63. https://doi.org/10.3233/NRE-192931

Appendix 1

CHILD NEUROPSYCHOLOGY, 2016
http://dx.doi.org/10.1080/09297049.2016.1191629

Routledge
Taylor & Francis Group

Body representation in children with hemiplegic cerebral palsy

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ABSTRACT

Clinical observations indicate that many children with hemiplegic cerebral palsy refrain from using or disregard the affected upper limb. The aim of the present study is to investigate deficits in different body representations (body schema, body structural description, and body image) in children with hemiplegic cerebral palsy (HCP) compared to typically-developing (TD) children. Three groups of children participated in this study: 42 TD children (aged 5.17–10.91 years), 23 children with right HCP (aged 5.83–10.92 years), and 22 children with left HCP (aged 5.67–10.90 years). The results demonstrate generalized deficits in all three body representations in children with HCP, and do not offer evidence for an effect of hemiplegia laterality.

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Hemiplegic cerebral palsy; Body representation; Body schema; Body structural description; Body image

Cerebral palsy (CP) is a condition caused by congenital or early non-progressive damage to the immature brain and comprises a group of disorders of movement and posture (Morris, 2007; Watkins & Rosenberg, 2002). A variety of motor disorders are associated with CP (e.g., spasticity, athetosis, and ataxia). Hemiplegic cerebral palsy (HCP), the subject of the present study, is characterized by unilateral spastic paresis or plegia attributable to a contralateral brain lesion (Mewasingh et al., 2004). Children with HCP show a delay in the acquisition of motor milestones and deficits in the organization of body movements of both the upper and lower limbs (Bax et al., 2005; Mewasingh et al., 2004). However, the modern definition of CP extends beyond motor deficits to encompass accompanying sensory and perceptual impairments, cognitive deficits, and learning and behavioral disorders (Bax et al., 2005; Morris, 2007; Rethlefsen, Ryan, & Kay, 2010).

Clinical observations indicate that children with HCP often disregard or do not use the affected upper limb in bimanual tasks, and fail to engage the hemiparetic limb as a support for the healthy one. This behavior is similar to the motor neglect observed in adults (Punt & Riddoch, 2006). The term "developmental disregard" has been proposed

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to characterize failure to use the affected arm and hand spontaneously in daily life (Houwink, Aarts, Geurts, & Steenbergen, 2011).

In clinical practice, children with HCP have commonly been observed as exhibiting functional alterations in the hemiparetic upper limb that resemble somatoagnosic or motor neglect disorders encountered in adults with acquired left hemiplegia. For example, these children fail to use the paretic hand as an assistive device for tasks such as tying shoelaces, or elect to use the mouth to remove the cap of a pen. In other circumstances, children ignore the affected superior limb so that it lies in a passive, sometimes awkward, dysfunctional position. Some children even sit on the paretic limb for several minutes at a time. In other cases, children develop negative attitudes towards the affected limb and complain about the inert limb or refer to it as a "thing". Occasionally, children refuse to use the paretic upper limb or remove the equivalent limb from toys.

Spatial neglect is also observed in HCP children in the clinical context (Katz, Cermak, & Shamir, 1998; Trauner, 2003). Children draw figures asymmetrically or neglect the side of the figure corresponding to the compromised domain. Trauner (2003) assessed hemispatial neglect in young children with early unilateral brain damage using two object removal preference tasks. The results suggest that spatial neglect, operationalized as less exploration of the objects in the contralateral hemispace, can be observed in young children after early unilateral brain damage.

According to Ajuriaguerra and Stucki (1969), some children with HCP appear to ignore or have no awareness of the affected limb and are therefore unable to use or even look at it. Ajuriaguerra and Stucki asserted that the functional deficits of the affected limb presented by children with HCP may not be entirely explained by motor difficulties, but rather hypothesized that perceptual-visual disorders exacerbate the motor disorders. Katz et al. (1998) suggested that unilateral neglect affects the assimilation of visual information from the environment. Houwink et al. (2011) interpreted disregard symptoms as a consequence of disuse, suggesting that attention deficits and lack of automatization may be aggravating factors. Alternatively, these observations can be interpreted as indications that body representation is impaired in some children with HCP. Therefore, in this study it is hypothesized that deficits in body representation compromise motor learning and motor use in daily activities.

Body knowledge constitutes an egocentric reference that allows an individual to function in his or her surrounding environment. Information on the configuration or perception of one's own body is provided by interoceptive, visual and somatosensory afferents and the monitoring of motor commands by means of proprioceptive feedback from motor execution (Murata & Ishida, 2007). The mental representation of the body is progressively developed over time by sensory afferents that maintain a link with motricity (Smith, 2001).

Based on the model of multiple sensory afferents, Sirigu, Grafman, Bressler, and Sunderland (1991) proposed the first systematic cognitive-neuropsychological description of body representation (see also Coslett, 2014). This model suggests that the processing of body-related knowledge includes at least three distinct domains of body representation: the body schema (BS), which supplies information regarding the present orientation of the body in space; the body structural description (BSD), which is a topological map of the human body; and the body image (BI), which provides lexical-semantic and visceral-affective information about the body.

Growing knowledge derived from functional neuroimaging and patient studies indicates that distinct cortical areas contribute to the implementation of three body representations (Berlucchi & Aglioti, 2010). The BS has been linked to the posterior parietal cortex, while the BSD and BI are associated with the ventral lateral occipito-temporal transition (extrastriate body area) and the insula, respectively.

The hemispheric laterality of representations is still unclear (Berlucchi & Aglioti, 2010). Disorders of the BSD such as autotopagnosia and finger agnosia are usually associated with left hemispheric lesions in adults (Buxbaum & Coslett, 2001). Motor hemineglect and disorders of body ownership, as observed in anosognosia for hemiplegia or hemiasomatognosia, are associated with right hemispheric lesions. The results of Trauner (2003) suggest that hemispheric localization bias is not necessarily observed in children with spatial hemineglect.

Body awareness—i.e., the perception of one's own body—is fundamental to motor control (Murata & Ishida, 2007). According to Heilman and Rothi (1997), the brain requires the assembly and maintenance of a motor program in order to track muscles and perform movements. Developmentally, somatosensory and motor afferences contribute to the development of mental representations of the body (Smith, 2001). During development, knowledge of the body influences the assembly of motor programs for new actions (Heilman & Rothi, 1997). Therefore, it is hypothesized that lesions of the immature brain, as in HCP, can produce disorders in body representation.

The purpose of this study is to investigate body representation deficits in children with congenital hemiplegia compared to typically-developing (TD) children. To this end, a battery of body representation tasks was employed that has been designed for children and adolescents with HCP (Fontes, Moura, & Haase, 2014). Given that this task battery was designed based on adult cognitive-neuropsychological studies (Coslett, 2014; Schwoebel & Coslett, 2005), we wanted to know more about its applicability in the context of developmental neuropsychology. We were also interested in characterizing the domain specificity of body representation deficits in HCP children. Previous data indicates that HCP children may present with very specific body representation deficits: for example, Frassinetti et al. (2012) observed that individuals with right unilateral brain damage presented with impairments in their own BSD, while individuals with left hemispheric lesions presented with impairments in the BSD of others. Accordingly, we investigated deficits in HCP children while taking into account the influences of age, intelligence, the domain of the body representations that is impaired, and laterality.

Method

Participants

The children in the HCP groups were recruited from a physical therapy outpatient facility at a large private university in a major urban area of south-east Brazil. The inclusion criteria consisted of a diagnosis of HCP, the absence of uncontrolled epilepsy, the ability to answer simple verbal commands, and regular attendance of a physical therapy program.

The children in the TD group were recruited from two public schools in the same region who regularly attended classes and had socio-demographic characteristics that are comparable to the HCP recruits. All research procedures were approved by the local advisory board (ETIC 250/09, COEP-UFMG) and participation in the study required written informed consent from parents and oral consent from the children.

The children recruited to participate in the study were divided into three groups as follows: 65 TD children, 24 children with right hemiplegic cerebral palsy (RHCP), and 25 children with left hemiplegic cerebral palsy (LHCP). To improve the power of the TD group as a control for the HCP groups, the intelligence range in the TD group was restricted within 1.0 standard deviation (*SD*) of the population mean in the Raven's Colored Progressive Matrices (CPM; Angelini, Alves, Custodio, Duarte, & Duarte, 1999) test, resulting in a final sample size of 42 children. In the HCP groups, 1 child in the RHCP group and 3 children in the LHCP group were excluded due to scoring more than 1.5 *SD*s below the population mean in the Raven's CPM test, leaving 23 and 22 participants in the RHCP and LHCP groups, respectively. The socio-demographic characteristics of the final sample are shown in Table 1. The final groups did not differ regarding age or gender (χ^2 = 1.27, p = .53). However, differences were observed in performance on the Raven's CPM test; in general, children from the HCP groups scored approximately 0.7 *SD*s below the TD group, but no differences were observed between the two HCP groups (Table 1).

Procedures

All children were evaluated in a quiet room at their school or rehabilitation center. For tests involving visual stimuli, children were seated on a chair in front of a desk with a laptop computer placed upon it. During the tests, the children were instructed to maintain the same position. The children were seated with their faces approximately 70 cm from the computer screen. For all tasks, accuracy was assessed and answers were coded as 1 (correct) or 0 (incorrect). No time limit was imposed, but the child was solicited to respond immediately after presentation. The final score was codified as the number of correct answers.

Instruments

Neuropsychological assessments included intelligence, basic sensorimotor tasks, and body representational tasks.

Intelligence

Intelligence was assessed by Raven's CPM (Angelini et al., 1999), which was applied to examine a possible association between intelligence and performance in body perception tasks.

Table 1. Demographic Characteristics.

	TD (n = 42)			RHCP (n = 23)			LHCP (n = 22)					TD × RHCP		TD × LHCP		RHCP × LHCP	
	n (%)	*M*	*SD*	*n* (%)	*M*	*SD*	*n* (%)	*M*	*SD*	$F(2, 84)$	*p* (*ANOVA*)	*p* LSD	*d*	*p* LSD	*d*	p LSD	*d*
Age (months)	42 (100)	94.40	20.79	23 (100)	104.09	17.93	22 (100)	97.82	20.04	1.76	.178	.064	0.50	.516	0.17	.294	0.33
Gender																	
Female	27 (64.30)	–	–	13 (56.50)	–	–	11 (50)	–	–	–	–	–	–	–	–	–	–
Male	15 (35.70)	–	–	10 (43.50)	–	–	11 (50)	–	–	–	–	–	–	–	–	–	–
Raven's CPM	42 (100)	0.29	0.53	23 (100)	−0.43	0.69	22 (100)	−0.36	0.63	14.04	.001	.001	1.17	.001	1.12	.698	0.11

Note. ANOVA = analysis of variance; d = Cohen's d; F = F statistic of ANOVA; LHCP = left hemiplegic cerebral palsy group; LSD = least significant difference; p = level of significance of post hoc test; p (ANOVA) = level of significance of ANOVA; RHCP = right hemiplegic cerebral palsy group; TD = typically-developing group.

Neuropsychological Sensorimotor Tasks

Hand, foot, and eye lateral dominance was assessed using the Laterality Task (Lefevre, 1972). Motor dexterity was examined using the Nine Hole Peg Test (9-HPT; Poole et al., 2005). Visual, auditory, and tactile extinction were clinically assessed using standard neurological examination procedures. Middle Finger Position Sense (recognition of the position of the joint) and Stereognosis (blindfolded recognition of common objects) tasks were examined in the upper limbs (Campbell, 2005).

Assessment of Body Representation

All tasks used to assess body representation were developed based on cognitive-neuropsychological (Sirigu et al., 1991) and neuroanatomical (Berlucchi & Aglioti, 2010) models developed for adults. The neuropsychological tasks for assessing body representation in the present study were developed in our laboratory. In order to explore all relevant domains of body representation, specific tasks for each level of representation (BS, BSD, and BI) were evaluated. The use of these tasks was verified in a previous study (Fontes et al., 2014). Internal consistency levels with the K-R20 formula were greater than 0.6 (Fontes et al., 2014).

Assessment of Body Schema (BS)

Hand Laterality Task

Pictures of single hands were presented on 10"×10" LCD screen and the child was instructed to indicate whether each picture was of a right hand or a left hand. This task was divided into two tests: the Oral Hand Laterality test, in which the child provided a verbal answer, and the Motor Hand Laterality test, in which the child raised the hand that corresponded to the stimulus. In the oral test, the child was verbally instructed to "say 'right' if the right hand is presented and 'left' if the left one is presented". In the motor test, the child was verbally instructed to "show the right hand if the right hand is presented, and show the left hand if the left one is presented". Twelve digital drawings of hands were presented in each test. The stimuli were six photographs of human hands in different positions (dorsal, palmar, and lateral rotated views of a hand with the fingers pointing medially or downward). The proportion of right- to left-hand trials was 6 to 6. Responses were coded as correct if the child correctly identified the hand verbally or by raising his or her corresponding hand.

Imitation of Meaningful Gestures Task

Stimuli for meaningful gesture imitation were presented on the computer screen and the child was instructed to correctly imitate the gesture, regardless of hand laterality. The child was verbally instructed to "correctly imitate the presented gesture". Meaningful gestures were ten animations: "waving goodbye", "asking for silence", "military salute", "pointing straight ahead", the "OK" sign, the "no" sign, "blowing a kiss", the "stop" sign, "clapping hands", and "pointing to one side". Representations in all trials were of the right upper limb/hand. Responses were coded as correct if the examiner considered the imitation to correspond to the model. The child was allowed to use either the non-plegic or paretic limb.

Imitation of Meaningless Gestures Task

Stimuli for meaningless gesture imitation were ten pictures: five pictures depicted fingers in specific positions, and five pictures displayed arbitrary positioning of the upper limb in relation to the trunk and head. As in the preceding task, items were presented on the computer screen and the child was instructed to correctly imitate the gesture, regardless of laterality. The child was verbally instructed to "correctly imitate the presented gesture". Responses were coded as correct if the examiner considered the imitation to correspond to the model. The child was allowed to use either the non-plegic or paretic limb.

Assessment of Body Structural Description (BSD)

Finger Gnosia Task

Finger gnosia was assessed using procedures previously described by Benton, Sivan, Hamsher, Varney, and Spreen (1994) and adapted by Dellatolas, Viguier, Deloche, and De Agostini (1998) (see also Costa et al., 2011). This 24-item task consisted of three parts: (a) with the hand visible, localization of single fingers touched by the examiner with the pointed end of a pencil (two trials on each hand); (b) with the hand hidden from view, localization of single fingers touched by the examiner (four trials on each hand); (c) with the hand hidden from view, localization of pairs of fingers simultaneously touched by the examiner (six trials on each hand). In accordance with Dellatolas et al. (1998), the children were given the choice of response method: they could name the touched fingers, point to the touched fingers on an outline drawing of the stimulated hand, or call out a number corresponding to the touched figure using a figure in which fingers were labeled one through five, beginning with the thumb. A correct answer was coded 1 and a wrong answer was coded 0. A total score ranging from 0 to 12 was calculated for each child. The internal consistency of this task is high (KR-20 = 0.79).

Visual Body-Part Localization Task

The child was shown a body part on a computer screen and instructed to point to the corresponding part of his or her own body. The child was verbally instructed as follows: "point on your own body to the same body part as the one presented". Visual stimuli were composed of pictures of twenty isolated body parts (hair, belly, foot, mouth, ankle, arm, leg, knee, neck, shoulder, face, nose, head, elbow, ear, chest, eye, hand, back, and wrist) presented in a pseudo-random order. Responses were coded as correct if the subject identified (pointed to) the correct body part, regardless of laterality.

Verbal Body Part Localization Task

The child was asked to point to a body part named by the examiner. The child was verbally instructed to "point on your body to the same body part as the one named". Verbal stimuli consisted of the names of eighteen body parts (hair, belly, foot, mouth, arm, leg, knee, neck, shoulder, face, nose, head, elbow, ear, chest, eye, hand, and back), which were spoken clearly by the examiner. Responses were coded as correct if the subject identified (pointed to) the correct body part, regardless of laterality.

Matching Body Parts by Location Task

The stimuli consisted of four body-part pictures simultaneously presented on the computer screen. One sample picture was presented above and the three test pictures were arranged in a row below. The child was asked to select from among the three pictures in the lower row the physical continuation of the single picture depicted above. The child was verbally instructed to "point to the picture of the body part which is nearer to or continues from the picture of [the body part in the sample picture]". For example, when the sample picture represented a leg, the response was correct if the child pointed to the picture of a foot (among foil images of an ear and a hand). The position of the correct choice in the lower row was pseudo-randomized. Eleven trials were carried out and responses were coded as correct if the subject identified (pointed to) the correct body part.

Assessment of Body Image (BI)

Matching Body Parts by Function Task

The stimuli consisted of four body-part pictures simultaneously presented on the computer screen. One sample picture was presented above and the three test pictures were arranged in a row below. The child was asked to select from among the three pictures in the lower row the body part that has a similar function or categorical relation to the body part depicted in the sample picture, or the body part that is "doing a similar thing". The child was verbally instructed "to point to or to say the name of the body part picture doing a similar thing to [the body part in the sample picture]". For example, when the sample picture represented a leg, the response was correct if the child pointed to or verbally indicated the picture of an arm (among foils of a shoulder and an ear). In another trial, when the sample picture represented an elbow, the response was correct if the child pointed to or verbally indicated the picture of a knee (among foils of a nose and a foot). Sample body parts were nine pictures of isolated body parts displayed in a pseudo-random order. Nine trials were carried out and responses were coded as correct if the subject identified (pointed to or verbally indicated) the correct body part.

Body Parts and Object Association Task

The stimuli consisted of four body-part pictures simultaneously presented on the computer screen. One sample picture was presented above and the three test pictures were arranged in a row below. The sample picture depicted a grooming tool or item of clothing or accessory, and the child was asked to select the matching body part from among the three pictures in the lower row. For example, in one trial the sample was a watch, and the correct response was the wrist; the foils were an ankle and an elbow. The child was verbally instructed to "point to or to say the name of the body part that is related to the [item in the sample picture]". Eleven trials were carried out and responses were coded as correct if the child identified (pointed to or verbally indicated) the matching body part.

Naming Body Parts Task

The child was asked to name eighteen isolated body parts, which were presented one at a time on the computer screen. The child was verbally instructed to "say the name of the presented body part". The same stimuli were used as those in the Verbal Body Part Localization task. Responses were coded as correct if the child correctly named the depicted body part.

Statistical Analyses

Possible associations between intelligence and age and task performance were examined using Pearson's bivariate correlation analysis. One-way analyses of variance (ANOVAs) were used to examine between-group differences in age and intelligence. For the remaining variables, group differences in accuracy were analyzed using analyses of covariance (ANCOVA) models, statistically controlling for intelligence, age, and the 9-HPT dominant hand. Pairwise post hoc analyses of group differences were conducted using least significant difference (LSD) tests. The risk of type I error was controlled by means of the LSD method. All statistical analyses were performed using the Statistical Package for Social Sciences (SPSS; SPSS Inc., 2009). Alpha was set to .05. Data are expressed as means ± *SD*s.

Results

Initially, a correlation matrix was calculated for all tasks (Table 2). First, we investigated whether or not the performance in the neuropsychological sensorimotor and body representation tasks is associated with age and intelligence.

The correlations between age and sensorimotor tasks are significant but low ($r < .34$) for the 9-HPT (dominant hand) and Stereognosis (dominant hand) tasks. The correlations between age and body representational tasks are significant but low ($r < .32$) for the Finger Gnosia (dominant and non-dominant hands), Matching Body Parts by Location, and Matching Body Parts by Function tasks (Table 2). The scores on all other sensorimotor and body representational tasks do not correlate with age.

Even though most of the correlations with age are low, we decided to explore in further detail the effect of age, as this characteristic shows large variability in the present sample. The distribution of scores for all tasks as a function of age was explored using scatterplots. Figure 1 shows the scatterplots of a representative task from each domain of body representation. Despite age variation, the children in the HCP groups always display lower levels of achievement in the variables of interest than the children in the TD group. Given the large variability of age in our sample, all further analyses use age as a covariate.

Several tasks correlate with Raven's CPM performance. In general, correlations are weak to moderate ($r = .26$ to $r = .44$, see Table 2). As intelligence differs between groups and correlates with task performance, all further analyses use the Raven's CPM score as a covariate.

Table 2. Correlation Coefficients.

	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21	22	23
1. Age (months)		−.23*	−.22*	.17	.01	.12	−.11	.18	.14	.34**	.01	.17	−.01	.18	.18	.32**	.28**	.08	−.06	.25*	.23*	.00	.14
2. Raven’s CPM			−.40**	−.41**	.16	.20	.05	.28**	.38**	−.03	.40**	.18	.12	.28**	.43**	.44**	.44**	.26*	.14	.35**	.15	.16	.36**
3. 9-HPT (dominant hand)				.38**	−.53**	−.54**	−.30**	−.51**	−.56**	−.34**	−.49**	−.28**	−.30**	−.23*	−.43**	−.51**	−.53**	−.34**	−.34**	−.52**	−.24*	−.37**	−.48**
4. 9-HPT (non-dominant hand)					−.06	−.05	−.08	−.20	−.44**	−.10	−.68**	−.14*	−.28**	−.30**	−.21	−.27*	−.43**	−.24*	−.25*	−.26*	.02	−.19	−.34**
5. Visual Extinction						.32**	.47**	.22*	.25**	.22**	.20	−.01	.15	.10	.18	.29**	.25*	.18	.26*	.24*	.24*	.19	.35**
6. Auditory Extinction							.19	.28**	.21*	.24**	.15	.08	.05	.02	.22*	.31**	.29**	.33**	.20	.23*	.13	.03	.28**
7. Tactile Extinction								.03	.17	−.01	.15	.12	.10	.04	.03	.14	.19	.00	.24*	.03	.03	.02	.17
8. Middle Finger Position (dominant hand)									.38**	.21*	.32**	.10	.27**	.25**	.18	.23**	.33**	.29	.12	.41**	.22**	.21*	.21
9. Middle Finger Position (non-dominant hand)										.30**	.60**	.21*	.26**	.39**	.51**	.54**	.59**	.41**	.30**	.50**	.23**	.28**	.53**
1. Stereognosis (dominant hand)											.27**	.13	.18	.21*	.20	.37**	.27**	.22**	.04	.37**	.18	.26**	.25**
11. Stereognosis (non-dominant hand)												.37**	.47**	.46**	.44**	.47**	.64**	.31**	.33**	.47**	.19	.27**	.59**
12. Oral Hand Laterality													.49**	.11	.29**	.30**	.43**	.09	.06	.15	.11	.32**	.29**
13. Motor Hand Laterality														.21	.36**	.29**	.44**	.13	.36**	.29**	.27*	.36**	.27*
14. Imitation of Meaningful Gestures															.39**	.35**	.35**	.42**	.24*	.43**	.31**	.22*	.41**
15. Imitation of Meaningless Gestures																.65**	.69**	.30**	.33**	.46**	.14	.37**	.50**
16. Finger Gnosia (dominant hand)																	.74**	.26*	.24*	.50**	.20	.29**	.54**

(*Continued*)

Table 2. (Continued).

	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21	22	23
17. Finger Gnosia (non-dominant hand)																		.27*	.34**	.54**	.20	.28**	.60**
18. Visual Body-Part Localization																			.32**	.33**	.15	.18	.47**
19. Verbal Body-Part Localization																				.27*	.25*	.16	.32**
2. Matching Body Parts, Location																					.34**	.25*	.55**
21. Matching Body Parts, Function																						.10	.21*
22. Body Part and Object Association																							.21*
23. Naming Body Parts																							

Note. **Correlation significant at the .01 level (2-tailed); *correlation significant at the .05 level (2-tailed).

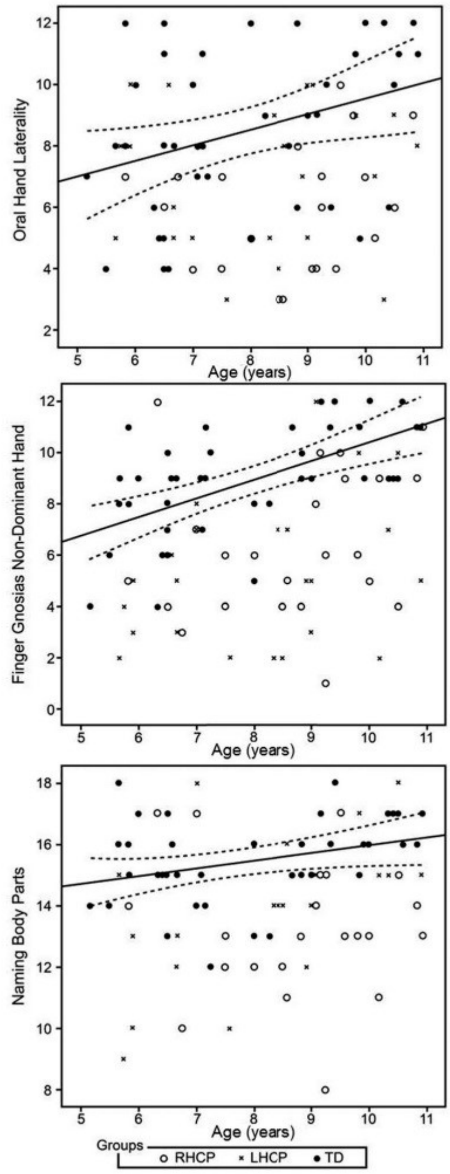

Figure 1. Scatter plots of the association between score and age relative to the mean performance of the TD group for (a) the Oral Hand Laterality task (linear $R^2 = .118$, $p = .026$), (b) the Finger Gnosia Non-Dominant Hand task (linear $R^2 = .349$, $p < .001$), and (c) the Naming Body Parts task (linear $R^2 = .097$, $p = .044$).
Note. LHCP = left hemiplegic cerebral palsy group; RHCP = right hemiplegic cerebral palsy group; TD = typically developing group. The solid line represents the line of best fit for the mean performance of the 42 TD children and the dashed lines indicate 95% confidence limits.

All tasks correlate with performance on the 9-HPT (dominant hand). In general, correlations are weak to moderate (r = .23 to r = .56, see Table 2). Thus, all further analyses use the 9-HPT (dominant hand) score as a covariate.

Some inter-correlations between body representation tasks are also significant. All BSD assessment tasks significantly correlate with two of the BS tasks (the Imitation of Meaningful Gestures and Imitation of Meaningless Gestures tasks, r = .24 to r = .69). All BI assessment tasks significantly correlate with two of the BS tasks (the Motor Hand Laterality and Imitation of Meaningful Gestures tasks, r = .22 to r = .41). The Naming Body Parts task, one of the BI assessments, significantly correlates with all BSD and BS assessment tasks (r = .27 to r = .60). Finally, some BI assessment tasks (the Matching Body Parts by Function and Body Part and Object Association tasks) significantly correlate with most BSD and BS assessment tasks (r = .22 to r = .37).

Neuropsychological Sensorimotor Tasks

A total of 40 children (95.2%) in the TD group exhibited a right-hand preference. The majority of children with HCP preferred to use the non-paretic hand and foot. Left-hand preference was observed in 20 (87.0%) of the children in the RHCP group, and right-hand dominance was observed in 22 (100.0%) of the children in the LHCP group. Right-foot laterality was observed in 38 (90.5%) of the children in the TD group. Left-foot preference was observed in 22 (95.7%) of the children in the RHCP, and right-foot dominance was observed in 21 (95.5%) of the children in the LHCP group. Right-eye laterality was observed in 24 (57.1%) of the children in the TD group. The majority of the children in the HCP groups preferred to use the eye on the non-paretic side; left-eye laterality was observed in 19 (82.6%) of the children in the RHCP group, and right-eye laterality was observed in 18 (81.8%) of the children in the LHCP group.

THe scores for the other neuropsychological sensorimotor tasks are shown in Table 3. Relative to the TD group, the two HCP groups were impaired with both hands in the motor dexterity task (9-HPT), but no differences are observed between the two HCP groups. No between-group differences are observed for the three extinction tasks. For the LHCP group, performance on the Middle Finger Position Sense task was significantly impaired for the both hands (the dominant and non-dominant hands). In the RHCP group, finger position sense was not significantly impaired for either hand, but the effect size is moderate to high (d = 0.66 for the dominant hand and d = 0.79 for the non-dominant hand). No differences in the Middle Finger Position Sense task are observed between the two HCP groups. In the Stereognosis task, the HPC groups are impaired in the non-dominant hand relative to the TD group. For the dominant hand, no between-group differences are observed. No differences in performance in the Stereognosis task are observed between the two HPC groups.

Body Schema (BS)

The results of the body-representational tasks are shown in Table 4. Significant between-group differences are observed for all tasks assessing BS. In the Oral Hand Laterality and Motor Hand Laterality tasks, both HPC groups displayed a significantly poorer performance than the TD group.

Table 3. Results of ANCOVAs for Neuropsychological Sensorimotor Tasks.

	TD (n = 42)		RHCP (n = 23)		LHCP (n = 22)		F (ANCOVA) [2,81]	p (ANCOVA)	η^2 partial (ANCOVA)	TD × RHCP		TD × LHCP		RHCP × LHCP	
	M	SD	M	SD	M	SD				p LSD	d	p LSD	d	p LSD	d
9-HPT (dominant hand)	23.28	3.93	31.00	10.85	29.46	4.77	8.03	.001	.16	.001	0.95	.009	1.41	.235	0.18
9-HPT (non-dominant hand)	25.55	3.78	339.96	403.68	381.20	393.62	4.55	.013	.10	.050	1.10	.004	1.28	.380	0.10
Visual Extinction	7.98	0.15	7.83	0.65	7.86	0.47	0.89	.413	.02	.202	0.32	.330	0.32	.707	0.07
Auditory Extinction	7.74	0.63	7.22	1.38	7.32	1.21	0.05	.954	.01	.771	0.49	.818	0.44	.939	0.08
Tactile Extinction	7.90	0.37	7.87	0.46	7.86	0.47	1.03	.362	.03	.157	0.08	.400	0.10	.512	0.01
Middle Finger Position (dominant hand)	7.52	0.99	6.43	2.11	5.73	1.91	4.02	.022	.09	.332	0.66	.007	1.18	.086	0.35
Middle Finger Position (non-dominant hand)	7.24	1.12	5.65	2.62	5.14	1.78	3.27	.043	.08	.244	0.79	.013	1.41	.197	0.23
Stereognosis (dominant hand)	3.67	0.53	3.52	0.95	3.45	0.74	0.36	.699	.01	.565	0.20	.408	0.34	.826	0.08
Stereognosis (non-dominant hand)	3.76	0.48	1.74	1.10	1.55	1.50	26.85	.001	.40	.001	2.38	.001	1.98	.480	0.14

Note. ANCOVA = analysis of covariance; d = Cohen's d; F = F statistic of ANCOVA; LHCP = left hemiplegic cerebral palsy group; LSD = least significant difference; η^2 partial = partial eta squared of ANCOVA; p = level of significance of post hoc test; p (ANCOVA) = level of significance of ANCOVA; RHCP = right hemiplegic cerebral palsy group; TD = typically-developing group.

Table 4. Results of ANCOVAs for Body Representation Tasks.

		TD (n = 42)		RHCP (n = 23)		LHCP (n = 22)		F ANCOVA [2,81]	p ANCOVA	η^2 partial ANCOVA	TD × RHCP		TD × LHCP		RHCP × LHCP	
		M	SD	M	SD	M	SD				p LSD	d	p LSD	d	p LSD	d
BS	Oral Hand Laterality	8.45	2.57	6.17	2.21	7.00	2.33	5.29	.007	.12	.002	0.95	.049	0.59	.166	0.36
	Motor Hand Laterality	9.90	1.95	7.35	2.84	6.73	2.41	13.72	.001	.25	.001	1.05	.001	1.45	.378	0.24
	Imitation, Meaningful Gestures	9.74	0.54	9.57	0.51	9.00	1.23	4.19	.019	.94	.830	0.33	.012	0.77	.022	0.60
	Imitation, Meaningless Gestures	8.48	1.38	7.09	2.13	6.41	2.09	3.74	.028	.09	.183	0.77	.008	1.17	.196	0.32
BSD	Finger Gnosia (dominant hand)	9.26	1.90	8.13	2.79	7.50	2.67	060	.551	.02	.760	0.47	.462	0.76	.295	0.23
	Finger Gnosia (non-dominant hand)	8.86	2.15	6.43	2.84	5.23	2.91	7.92	.001	.16	.034	0.96	.001	1.42	.096	0.42
	Visual Body-Part Localization	19.52	0.74	18.61	1.75	18.82	1.47	1.06	.351	.03	.159	0.68	.314	0.61	.630	0.13
	Verbal Body-Part Localization	17.90	0.30	17.35	0.71	17.32	1.04	3.71	.029	.08	.042	1.02	.011	0.77	.686	0.03
	Matching Body Parts, Location	9.90	0.98	8.87	1.63	8.77	1.23	2.07	.133	.05	.122	0.77	.057	1.02	.783	0.07
BI	Matching Body Parts, Function	7.45	1.37	6.78	1.59	6.59	1.59	1.80	.171	.04	.138	0.45	.080	0.58	.850	0.12
	Body Part and Object Association	10.88	0.40	10.74	0.75	10.50	1.10	1.28	.258	.03	.322	0.24	.586	0.46	.116	0.24
	Naming Body Parts	15.43	1.38	13.26	2.20	14.05	2.40	3.31	.042	.08	.012	1.18	.203	0.71	.159	0.34

Note. ANCOVA = analysis of covariance; d = Cohen's d; F = F statistic of ANCOVA; LHCP = left hemiplegic cerebral palsy group; LSD = least significant difference; η^2 partial = partial eta squared of ANCOVA; p = level of significance of post hoc test; p (ANCOVA) = level of significance of ANCOVA; RHCP = right hemiplegic cerebral palsy group; TD = typically-developing group.

In the Imitation of Meaningful Gestures and Imitation of Meaningless Gestures tasks, only the LHCP group was impaired relative to the TD group, but effect sizes for the differences between the TD and RHCP groups are non-negligible (d = 0.33 and d = 0.77, respectively). Significant between-group differences for the HPC groups are only observed in the Imitation of Meaningful Gestures task, where the LHCP group was impaired relative to the RHCP group.

Body Structural Description (BSD)

In the BSD assessments, significant between-group differences are observed for the Finger Gnosia (non-dominant hand) and Verbal Body Part Localization tasks. Finger Gnosia was consistently impaired for the non-dominant hand in both HPC groups relative to the TD group. Performance in the Finger Gnosia (dominant hand) task was poorer in the RHCP (d = 0.47) and LHCP (d = 0.76) groups, but statistically insignificant. Both HPC groups were significantly impaired relative to the TD group in the Verbal Body Part Localization task.

No significant between-group differences were found for the Visual Body Part Localization and Matching Body Part by Location tasks. However, effect sizes for the post hoc comparisons between each HPC group and the TD group are in the moderate range (Visual Body Part Localization: d = 0.68 for RHCP vs. TD, d = 0.61 for LHCP vs. TD; Matching Body Part by Location: d = 0.77 for RHCP vs. TD, d = 1.02 for LHCP vs. TD). No statistically significant differences are observed between the two HPC groups for the BSD tasks.

Body Image (BI)

Significant between-group differences in BI tasks are observed only for the Naming Body Parts task, where the RHCP group was impaired relative to the TD group. However, the effect size for the difference between the LHCP and TD groups is in the moderate range (d = 0.71).

No significant differences and moderate effect sizes are observed for the comparison between groups in the two remaining tasks assessing BI (Matching Body Parts by Function and Body Parts and Object Association). Performance in the Matching Body Parts by Function task was lower in the RHCP (d = 0.45) and LHCP (d = 0.58) groups, although these differences are not statistically significant. A similar pattern is observed for the Body Parts and Object Association task, where both the RHCP (d = 0.24) and LHCP (d = 0.46) groups exhibit scores that are lower but not statistically different from those of the TD group. No statistically significant differences are observed between the two HPC groups in the BI tasks.

Discussion

In this study, we comparatively investigated different forms of body representation in groups of children with right and left HCP and TD children. For this purpose, we used a previously validated set of experimental tasks (Fontes et al., 2014). Neuropsychological assessment evaluated three types of body representation (BS, BSD, and BI) in accordance with current cognitive-neuropsychological models of body representation in adults (Berlucchi & Aglioti, 2010; Coslett, 2014; Sirigu et al., 1991).

The main finding of this study is that disorders of body perception and representation occur in children with HCP. In the Oral Hand Laterality, Motor Hand Laterality, Finger Gnosia (non-dominant hand), and Verbal Body Part Localization tasks, both HPC groups demonstrated impaired performance relative to the TD group. In the Imitation of Meaningful Gestures and Imitation of Meaningless Gestures tasks, only the LHCP group was impaired relative to the TD group, but the effect size of the difference between the TD and RHCP groups is non-negligible. In the Naming Body Parts task, only the RHCP group showed a slightly impaired performance relative to the TD group, but the effect size of the difference between the TD and LHCP groups is in the moderate range. Significant differences between the RHCP and LHCP groups are observed only in the Imitation of Meaningful Gestures task, with lower performance in the LHCP group. All other tasks have not yielded significant between-group differences, but effect sizes for comparisons between the HCP groups and the TD group are substantial.

These results indicate that HCP children perform poorly across several body representational tasks compared to TD children. In the following sections, the results will be discussed with respect to the effects of age, intelligence, the impaired domain of body representation, and impairment laterality.

Results Relating to the Effects of Age

In this study, the observation of a weak correlation between age and body representational tasks suggests that the body representations under assessment are already well developed in the age range of the study participants. From the perspective of cognitive neuropsychology, the three distinct body representation levels (BS, BSD, and BI) develop from infancy to adulthood (Camões-Costa, Erjavec, & Horne, 2011).

The BS, also known as dynamic representation of the relative position of body parts, is functional from at least 3 months of age, when a baby becomes aware of his or her own body as a dynamic and organized entity (for a review, see Rochat, 2010). Rochat (1998) conducted a series of experiments in which different views of babies' own legs were presented to them. It was observed that, from 3 months of age, babies participate in the exploration of their own body, both perceptually and actively.

Funk, Brugger, and Wilkening (2005) applied the hand laterality judgment task to 20 TD children and found that children of preschool age (5–6 years) are able to perform mental rotation and right-left discrimination. The performance of all children in this study was greater than 50%: 12 out of 20 children performed above 60.41% (mean age = 6.70 years, *SD* = 0.50 years). By comparison, in the present study, the success rate of the TD group (mean age = 7.87 years, *SD* = 1.73) is 70.42% and 82.50% for the Oral Hand Laterality and Motor Hand Laterality tasks, respectively. The high success rates observed in our study can be explained by the fact that the children assessed were older than those assessed in Funk et al. . The results of Funk et al. also indicate that changes in hand position during the task have a significant effect on performance, suggesting that, unlike motor imagery in adults, motor imagery in young children is driven by motor processes. The fact that HCP children have motor changes arising from injury to the immature brain may explain the lower performance level of the HCP children in our study.

In a similar study, Caeyenberghs, Tsoupas, Wilson, and Smits-Engelsman (2009) evaluated hand laterality judgment in 58 TDE children, divided into three different age groups: 7–8 years, 9–10 years, and 11–12 years. Their results show that the reaction time in mental rotation tasks for all groups increased with rotation angle, indicating that children between the ages of 7 and 12 years are able to perform mental rotation tasks. However, it was found that younger children had a lower success rate and longer reaction times. The authors further showed that at approximately 11–12 years of age, children perform similarly to adults (Caeyenberghs et al., 2009).

The BSD, also known as visuospatial body knowledge, is derived from the child's early experiences with his or her own body, such as getting dressed, undressing, and playing games that provide visual information related to the location of body parts (Slaughter, Heron, Jenkins, & Tilse, 2004). The experiments of Slaughter and colleagues (Slaughter et al., 2004; Slaughter, Heron-Delaney, & Christie, 2012; Slaughter, Heron, & Sim, 2002) found that children between the ages of 15 and 18 months are able to visually discriminate human body parts, suggesting that infants have a visuospatial representation of the body after the first year of life.

Brownell, Nichols, Svetlova, Zerwas, and Ramani (2010) studied the progress of topographical characteristics of the body in 61 children between the ages of 20 and 30 months through the application of five body self-awareness assessment tasks. They observed that children have an explicit but rudimentary topographical representation of their own bodies at 2.5 years of age, and that this representation continues to develop throughout the preschool period. The authors also found that, during the second to third year of life, awareness of the topographic or spatial organization of the body itself also appears.

Witt, Cermak, and Coster (1990) studied the development of body-part identification in 113 TD children between the ages of 11 and 25 months. The children were asked to point to 20 body parts on a doll. These authors found that only a minority of 12-month-old infants were unable to accurately locate body parts. At the end of the study, the authors concluded that the ability to locate body parts increases progressively with age. According to Slaughter et al. (2004), infants acquire the ability to identify their own body parts between 12 months and 4 years of age, and that this developmental sequence is quite regular in all children.

BI, also known as semantic body knowledge, is clearly related to the development of lexical comprehension and production that enables children to correctly identify and designate body parts. According to Camões-Costa et al. (2011), the lexical knowledge of different body parts gradually increases between the first and third years of life. The authors also suggest that the development of the ability to name body parts is highly related to the topographic representation of the body (see also Auclair & Jambaqué, 2014).

According to Gallahue (1982) and Simons and Dedroog (2009), the ability to name and identify body parts begins to develop at 3 years of age and is completely developed at 5 years. Simons and Dedroog applied four "naming and pointing" body-part tasks, similar to the Naming Body Parts and Visual Body Parts tasks used in this study. The performance of the 124 children of 3–6 years of age in the control group was 64.88%

and 59.82% for the pointing and naming tasks, respectively. In the present study, the performance of the 42 children 5.17–10.91 years of age in the TD group was 97.60% and 85.72% for the Visual Body Parts and Naming Body Parts tasks, respectively.

According to Facon, Facon-Bollengier, and Grubar (2002), older children are more experienced and therefore more likely to expand their vocabulary. Vallaey and Vandroemme (1995) suggested that the development of language is very closely related to the development of body awareness. According to Gallahue (1982), children at 3 years of age are able to identify and name only the most important parts of the head, torso, and limbs, whereas at 4 years, children can identify and name a larger number of parts of the head, torso, and limbs, and at 5 years, children can identify and name almost all parts of the head, torso, and limbs. In a study by Simons and Dedroog (2009), TD children of 3–6 years of age were able to point to and name these same parts of the body.

Brownell et al. (2010) found a significant correlation between locating body parts (BSD) and gesture imitation tasks (r = .53, p < .001). Camões-Costa et al. (2011) observed that the development of the ability to name body parts (BI) is highly correlated with the topographic representation of the body (BSD). According to Auclair and Jambaqué (2014), development of a visuospatial representation of the body influences lexico-semantic processing. Auclair and Jambaqué also stressed the importance of the action's role in the development of body representation and semantic organization.

For Müller, Sokol, and Overton (1998), the BSD gradually emerges from the BS, and the development of BI gradually emerges from the BSD. In accordance with this hypothesis, our study also found significant correlations (see Table 2) between body representation assessment tasks: 1) all BSD assessment tasks significantly correlate with the Imitation of Meaningful Gestures and Imitation of Meaningless Gestures tasks (BS); 2) all BI assessment tasks significantly correlate with the Motor Hand Laterality and Imitation of Meaningful Gestures tasks (BS); 3) the Naming Body Parts task (BI assessment) significantly correlates with all BSD and BS assessment tasks; and 4) other BI assessment tasks (Matching Body Parts by Function and Body Part and Object Association) significantly correlate with most BSD and BS assessment tasks.

Results Relating to the Effects of Intelligence

Although the children with HCP did not exhibit intelligence deficits in our study, both HCP groups displayed significantly lower performance on Raven's CPM relative to the TD group (d > 1.12). Furthermore, intelligence significantly correlates with five out of nine neuropsychological sensorimotor tasks and seven out to twelve body representation tasks. Cognitive deficits in HCP have been a recurring problem in neuropsychological studies because intelligence in HCP groups is frequently lower than that of controls (Levine, Kraus, Alexander, Suriyakham, & Huttenlocher, 2005; Muter, Taylor, & Vargha-Khadem, 1997; Trauner, Nass, & Ballantyne, 2001). Many studies have also observed considerable effect sizes when comparing the mean intelligence of children with HCP and TD children (Ashcraft, Yamashita, & Aram, 1992; Muter et al., 1997; Stiles, Trauner, Engel, & Nass, 1997; Thevenot et al., 2014; Tillema et al., 2008; Trauner, 2003; Trauner et al., 2001). Thus, as we cannot completely rule out the possibility of a relationship between intelligence and difficulties with body representation, we carefully controlled for the effects of intelligence in our study.

Results Relating to the Impaired Body Representation Domain

The results of our group analyses did not identify the existence of a specific form or representational domain that was affected to a larger extent in the children with HCP. Therefore, we suggest that the difficulties presented by children with HCP in the three body representations may reflect more general effects of damage and repair to the developing brain. Clinically observed symptoms in HCP children are quite similar to somatoagnosic or motor hemineglect, but may be associated with more general developmental difficulties rather than specific neural lesions of the central nervous system.

A study by Bertoldi, Ladewig, and Israel (2007) found that children 7–10 years of age with motor deficiencies (CP and myelomeningocele) showed good performance in identification of parts of their own body (the Visual Body Parts task) but had difficulty in recognizing the movement of parts of the body (kinesthetic perception) and focusing their attention on the body parts of other people. These data confirm the inadequate development of body awareness in HCP children. Studies by Simons and colleagues (Simons & Dedroog, 2009; Simons, Leitschuh, Raymaekers, & Vandenbussche, 2011) found that children with mental retardation and/or psychiatric disorders showed significantly lower performance than the control group in pointing (BSD) and naming (BI) body-part tasks. According to Christie and Slaughter (2009), in order to carry out such tasks, some lexical-semantic and visuospatial knowledge of the body is needed because the child must locate the named body part. Thus, success in the task depends on an intact topographical representation. Accordingly, the significant correlations that were found in the present study among tasks assessing the three distinct representational domains are justified in children with HCP.

Results Relating to the Effects of Impairment Laterality

Unlike patterns observed in adults with brain injury, we did not observe specific effects of the laterality of hemiplegia on disorders of body perception and representation in this study. According to Vossel et al. (2011) and Karnath and Rorden (2012), unilateral neglect syndrome in adults is more often found in patients with right temporoparietal damage, e.g., individuals with left hemiplegia. According to Berlucchi and Aglioti (2010), the cortical regions related to the body are present in both brain hemispheres. However, several studies of brain injury in adults (Peelen, Atkinson, Andersson, & Vuilleumier, 2007; Schwarzlose, Baker, & Kanwisher, 2005) have provided evidence of strong right-hemisphere dominance.

Chaminade, Meltzoff, and Decety (2005) used functional magnetic resonance imaging to evaluate brain activity in healthy subjects during imitation of gestures tasks and concluded that the BS is related to areas in the bilateral parietal gyrus. However, actions that involve visual perception are predominantly related to areas in the right hemisphere because important aspects of visual attention are necessary to encode a gesture within a space.

Studies of children with HCP have not yet provided clear evidence as to the laterality of impaired body representations. Katz et al. (1998) assessed children with HCP and found no evidence that children with right hemispheric damage had left unilateral neglect but did identify greater attention and perception deficits in children with right hemispheric lesions (left hemiplegia) than in children with left hemispheric lesions (right hemiplegia). In general, in our study, the scores of the LHCP group are lower than the scores of the RHCP group. However, in the Oral Hand Laterality and Naming of Body Parts tasks a slightly better performance is observed in the LHCP group. Thal et al. (1991) found that children with left hemispheric damage (right hemiplegia) are more impaired in expressive language than those with right hemisphere damage (left hemiplegia), while children with right hemispheric damage have more receptive delays than the left hemispheric damage group.

Kolk and Talvik (2002) assessed cognitive performance in children with handedness ipsilateral to a brain lesion and revealed that the children with right hemispheric lesions scored significantly lower than the children with left hemisphere lesions on tasks involving attention, sustained concentration, executive function, and the visuospatial domain. Conversely, performance on visuomotor precision tasks was significantly lower in the children with left hemispheric lesions than in the children with right hemispheric lesions. In this study, the authors did not find significant differences between the performances of the children with right and left hemisphere lesions in language tests or motor and sensory integration abilities.

Other studies have observed greater impairments in children and adolescents with right hemiplegia. Mutsaarts, Steenbergen, and Bekkering (2007) applied two mental rotation tasks to 19 adolescents with HCP (11 with right hemiplegia and 8 with left hemiplegia; mean age = 16.20 years, *SD* = 2.00 years) and 9 TD controls and noted that simulation of internal movements is impaired in individuals with right hemiplegia. Steenbergen, Meulenbroek, and Rosenbaum (2004) examined the differential roles of both hemispheres in motor planning by comparing the performance of adolescents with HCP in two tasks of object manipulation. Participants with right hemiplegia showed anticipatory planning problems (e.g., not selecting a favorable initial grip to complete the task in a comfortable posture), while left hemiplegia participants showed relatively unaffected anticipatory planning. A study by Elk et al. (2010) investigated the functional and neural dynamics of motor imagery in hand laterality judgment tasks of 10 adolescents with right hemiplegia (mean age 18.30 years, *SD* = 1.20 years) and 10 TD left-handed adolescents (mean age 19.70 years, *SD* = 2.20 years). Based on reaction times and electroencephalogram (EEG) analyses, it was observed that the participants with right hemiplegia had an impaired ability to use motor imagery. Unlike the present results, which show no evidence of lateral dominance, these studies provide evidence for greater deficits in children and adolescents with right hemiplegia. The present results agree with those of Kirkpatrick, Pearse, Eyre, and Basu (2013), who were unable to find any significant motor planning differences between children with right and left congenital hemiplegia. This result could be attributed to the heterogeneity of designs, samples and measures used in the different studies. The question of interhemispheric specialization of body representation is also not settled in the adult literature (Braun, Desjardins, Gaudelet, & Guimond, 2007).

Hand dominance was not always non-paretic in the RHCP group. It is only possible to conjecture that hand laterality could be genetically influenced (Ooki, 2014), or that the degree of motor impairment varies from child to child and that some children may not develop a preference for the unimpaired hand. No child was being treated with constraint-induced movement therapy, so this result cannot be attributed to intervention effects.

Final Considerations and Conclusions

The results of this study demonstrate the existence of disorders of perception and body representation in children with HCP at different body representation levels. The present data support the initial hypothesis that a lesion in the immature brain, such as in HCP, is associated with generalized disorders of body representation. Although the sample size of the present study is small, it is important to note that attempts were made to recruit all available and capable (of an eligible age and capable of responding to tasks) children for evaluation. Additionally, we wish to highlight the consistency of the results: even when the results are not statistically significant, the effects always trend in the same direction, which has significance for the interpretation and reliability of the data.

The present study yields several original and pioneering findings for the field of neuropsychology. A low correlation between age and body representation assessment tasks suggests that the representational domains examined (BS, BSD, and BI) were already well developed in the age range of the participants (5–11 years). The study importantly addresses the issue of intelligence (i.e., here and in other studies, intelligence was measured as being lower in the HCP groups than in the TD group) by implementing careful statistical controls to ensure a robust analysis. Additionally, significant correlations between body representation assessment tasks confirmed hypothesized interactions between the different domains of perception and representation of the body (see Sirigu et al., 1991). The present results have not identified a specific form or representational domain that is preferentially affected in children with HCP, which leads to the conjecture that general developmental difficulties underlie motor deficits in these children. Furthermore, an effect of hemiplegia laterality was not observed, and research has yet to provide clear evidence of the laterality of body representation impairments in children with HCP. It is therefore concluded that children with HCP experience laterality-independent, generalized impairments in all body representational domains.

Finally, this work has practical implications for the clinical rehabilitation of children with HCP. Because perceptual changes related to the body are easily detected in clinical practice, we emphasize the need to evaluate these parameters and, importantly, verify whether functional independence (e.g., activities of daily living and school learning) and rehabilitation can be influenced by the presence of disorders in body perception and representation. Future studies focusing on the interaction of body perception disorders and rehabilitation will further inform the clinical approach to HCP.

Disclosure Statement

No potential conflict of interest was reported by the authors.

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Appendix 2

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ORIGINAL RESEARCH
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Motor Imagery Development in Children: Changes in Speed and Accuracy With Increasing Age

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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, 8–9, 10–11, and 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 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

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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 (1, 2). According to Jeannerod (3) 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 (3, 4). Thus, MI exhibits many of the properties of motor planning and is considered a valid method for training the internal action control model (5). The internal motor control model proposed by Wolpert (4) 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 (4, 6). 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 (6, 7).

According to Jeannerod (8), 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 (9–12). 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 (2, 13, 14). Indeed, improvements in the performance of motor skills associated with MI training have been documented in healthy people (15, 16) and in clinical populations, particularly in post-stroke patients (17). 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 (18).

To improve motor skills, individuals must imagine all the sensations that accompany the physical performance of the imagined task (19). 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 (20–23). 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 (23). In a recent study involving transcranial magnetic stimulation (TMS), Hyde et al. (24) suggest that the motor cortex is activated during the performance of 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 (25) was confirmed by studies showing that biomechanical constraints that apply to physical motion also restrict imagined motion (21). 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 (1, 26). de Lange et al. (27) evaluated brain activation of healthy individuals while performing the mental rotation task using functional magnetic resonance 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 (19, 25, 28). 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 (27). 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 (29). However, there is great controversy as to the minimum age when a child is able to engage in tasks using MI (1, 20, 22, 30, 31). Moreover, the age when they reach development comparable to that observed in adults remains unclear. According to Funk et al. (32), 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 (33–37).

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 (20, 21, 32, 38), suggesting that in this age group they are already capable of performing MI based on motor processes. In the study by Funk et al. (32), 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. (22) 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. (32) 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 (39).

Caeyenberghs et al. (21) 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. (38) 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. (20) 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 to 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 vs. 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, 8–9, 10–11, 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 (40). 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.

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.

Right–Left Orientation

To evaluate right-left orientation we used the Right-Left orientation test (41). 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 (27). 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 (0°, 90°, 180°, and 270°). 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 (1, 25). This effect is characterized by an increase in RT as a function of the rotation angle of the stimuli (17). 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 90° (medial rotation) is faster than when the right hand at 90° is shown (25, 27).

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

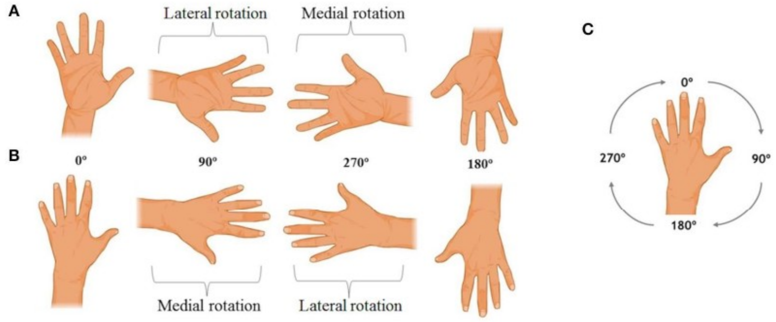

FIGURE 1 | Examples of the hand laterality judgment task stimuli. In **(A)**, the right hand stimuli are observed in palm view. In **(B)**, the left hand stimuli are presented in back view, and in **(C)** the rotation direction of the task stimuli is indicated.

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.

Effects of Biomechanical Constraints

Medial Rotation vs. 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 and 8–9 years of significantly slower than the groups 10–11, 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 vs. 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, 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

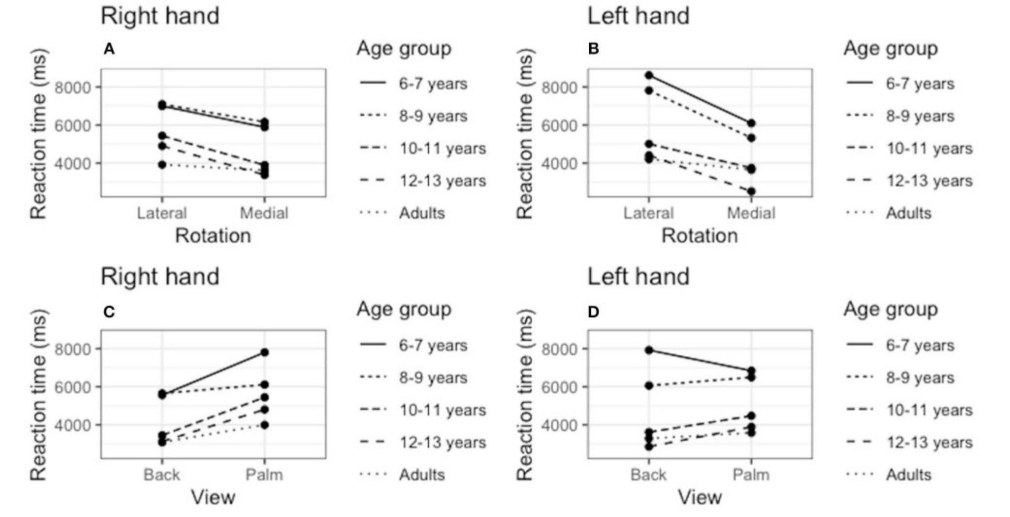

FIGURE 2 | Effect of biomechanical constraints. We verified the reaction time (RT) averages to judge the stimuli in medial rotations compared to 'lateral rotations for the right hand **(A)** and left hand **(B)**. We also compared the RT to judge the stimuli presented in the palmar and dorsal views for both the right **(C)** and left **(D)** hands.

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).

Age Differences

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

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 [$F_{(4, 180)} = 17.560$; $p < 0.00$; $\eta^2 = 3.79$]. The groups 6–7 and 8–9 years were significantly less accurate than the groups 10–11, 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 10–11, 12–13 years, and adult responded similarly in terms of accuracy ($p > 0.05$).

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, 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 and 12–13 years).

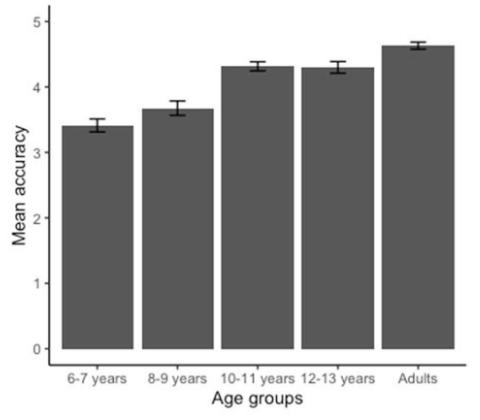

FIGURE 3 | Mean values obtained for the accuracy of the hand laterality judgment task. Groups 6–7 and 8–9 years were less accurate than groups 10–11, 12–13 years, and adult ($p < 0.001$). Error bars indicate standard error.

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,

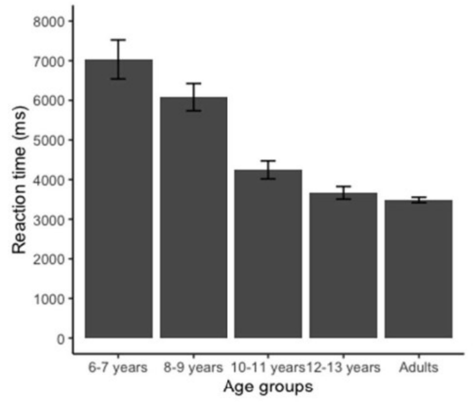

FIGURE 4 | Reaction time (RT) for the hand laterality judgment task. Group 6–7 years presented longer RT than groups 8–9, 10–11, 12–13 years, and adult ($p < 0.001$). Group 8–9 years showed longer RT than groups 10–11, 12–13 years, and adult ($p < 0.001$). Error bars indicate standard error.

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 (32, 43, 44). According to Belmont and Birch (45), 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 (21, 46, 47). 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. (20). After analyzing some studies, Spruijt et al. (20) 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 (48, 49).

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 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. (30) 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. (30) 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 (1, 22, 27). 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 (1). 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 (50). For Parsons (25), 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. (22) 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 (19, 25). 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 et al. (51) 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 et al. (52), 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 (52), 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 (51).

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 (1, 20, 21, 39). The study by Caeyenberghs et al. (21) compared the performance in MI through the HLJ task of 7 and 8 year old, 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. (20) 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 (21, 22, 53).

Our results point to an improvement in MI capacity as age increases. Similar results were also found by Caeyenberghs et al. (21). This improvement in MI as age is supported by the development and maturation of a set of complex cognitive processes (21). Significant structural and functional changes occur in the child's brain during childhood. According to Casey et al. (52) 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. (52) 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. (38). 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 (21, 38), 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. (38) 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 (20, 21, 54, 55). 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 (56). 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 (56). Vargas et al. (57) 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 (20, 21, 54, 55). According to previous studies, motor planning ability and motor skills may also influence MI performance (37, 38). 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 (58). 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. (59) 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.

DATA AVAILABILITY STATEMENT

The datasets generated for this study are available on request to the corresponding author.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Research Ethics Committee of the Federal University of Minas Gerais (COEP/UFMG). Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

DS and TC delineated the study. PF, RB, and VH conducted the neuropsychological evaluation. All authors contributed in analyzing the results and writing the paper. All authors read the final version of the paper and agree with the content of the manuscript.

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Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Appendix 3

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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

1. Introduction

The ability to perform various hand activities is reduced in children with hemiplegic cerebral palsy (HCP). Sensory and motor impairments observed in the affected upper extremities are a major cause of functional compromise (Rosenbaum et al., 2006).

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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.

2. Methods

2.1. 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.

2.2. 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).

2.3. 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.

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

2.4. Interventions

2.4.1. 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 1080s.

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.

2.4.2. 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.

2.5. 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.

2.6. 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).

3. 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

Table 2
Characteristics of study participants

Characteristics	Intervention group (n = 12)	Control group (n = 12)		
	%	%	χ2	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.

Table 3
Between-group comparison of intelligence and working memory

Intelligence	Intervention group M (sd)	Control group 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.

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

	Pre-test	Post-test	Follow-up	ANCOVA			*Post hoc*		
	M (sp)	M (sp)	M (sp)	*F*	*P*	η2	Pre-test x Post-test	Pre-test 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.

results of between-group comparisons are provided in Table 2.

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; $p < 0.980$).

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; $p < 0{,}001$; $\eta 2 = 0.254$), 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 ($p > 0.05$).

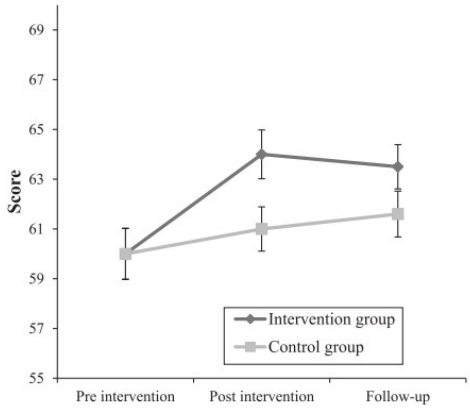

Fig. 1. Progression of the evaluation means carried out at three different time points. Note: Each point represents the average obtained by the groups in the AHA test during pre-intervention, post-intervention and follow-up measurements.

4. 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 $\alpha = 0.05$ 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.

4.1. 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).

5. 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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Conflict of interest

The authors report no conflicts of interest.

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