MIRROR (A)SYMMETRY? VISUO-PROPRIOCEPTIVE INTERACTIONS IN INDIVIDUALS WITH SPASTIC HEMIPARETIC CEREBRAL PALSY

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Mirror (a)symmetry? Visuo-proprioceptive interactions in individuals with spastic hemiparetic cerebral palsy

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Abstract

The work presented in this thesis aimed to get more insight into the previously reported positive effects of mirror visual feedback in children with spastic hemiparetic cerebral palsy (SHCP) and into visuo-proprioceptive interactions in children and adolescents with SHCP during goal-directed matching tasks. Individuals with SHCP have unilateral motor impairments that hamper them in accurate movement performance. In conjunction with the motor problems, these individuals experience sensory problems. The first study in this thesis (chapter two) found that mirror visual feedback of the impaired arm in SHCP led to significantly higher levels of neuromuscular activity than mirror visual feedback of the less-impaired arm. This indicates that the mirror-effect was not just caused by the illusory perception of symmetry between two limbs, and confirmed that the beneficial effect is dependent on mirror visual feedback of the less-impaired arm. In chapter three and four it was demonstrated that the ability of children with SCHP to match one (matching) hand with the position of the other (reference) hand, without visual information, is deteriorated when compared to typically developing children. However, if visual information of the static reference arm was available to the participants, the matching accuracy of the matching hand was significantly higher. Mirror visual feedback of the reference arm, generated by placing a mirror in between the arms in the sagittal plane, created the illusion that both hands were already at the endpoint. However, this did not impact upon the matching accuracy of the matching arm and resulted in similar error scores as regular feedback of the reference arm. Chapter five showed that moving the less-impaired arm in synchrony with the impaired arm resulted in higher matching accuracy than moving the impaired arm alone. Moreover, mirror visual feedback of the less-impaired arm improved matching accuracy for a subset of the participants. The effects of a short practice of a bimanual matching task with (mirror) visual feedback of the less-impaired arm on matching accuracy of the impaired arm was studied in chapter six. The results showed a higher matching accuracy of the impaired arm after the practice period. However, the role of the mirror is still inconclusive in this respect. From this it can be concluded that for individuals with SHCP practice of a matching movement can induce a transfer from visual to proprioceptive control of movement. Taken together, the work in this thesis showed that the deficit in position sense of the impaired arm in individuals with SHCP can be modified by visual feedback of the less-impaired arm. Although the role of mirror visual feedback is still inconclusive, it seems that motor learning can induce a transfer from visual to proprioceptive control of movement, which can have implications for therapy.

Author’s declaration

The work in this thesis was carried out in accordance with the regulations of the Manchester Metropolitan University. The work is original and no part of the thesis has been submitted for any other academic award. The following assistance was received in completing the work:

- During data collection I was assisted by four Master students. Anniek Geerlings and Marjolein Smit assisted in the data collection of chapters two, three, four, and five. Monika Zakrocka assisted in the data collection of chapters three and four. They assisted by starting and stopping the measurement recordings on the computer. In addition, they performed all measurements on the typically developing children. Anniek Geerlings (physiotherapist) measured the Tardieu Score and taught me how to measure it. Wietske Waterlander assisted in the data collection of chapter five by starting and stopping the measurement recordings on the computer.

- Siro Otten and Hans Agricola (VU University Amsterdam) assisted in the construction of the experimental setup.

Any views expressed in this thesis are those of the author and do not represent those of Manchester Metropolitan University.

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Chapter 1

Introduction

Introduction

In daily life, we use our upper limbs for almost every movement and they are therefore extremely important for functional independence. The importance of our arms for everyday life is especially highlighted when one cannot use his/her arms due to e.g. a particular motor disorder. This is the case in children with spastic hemiparetic cerebral palsy (SHCP). Due to brain damage during early development these children have motor disorders (i.e. loss of motor function) on one side of the body (i.e. one arm and one leg; Bax et al., 2005; Miller, 2007). As a result of this unilateral impairment these children experience problems with the performance of daily movements, predominantly of movements that require the involvement of both arms, which severely hampers their capacities and functional independence. We can thus state that adequate control of both hands is essential for everyday movement performance. Another vital factor for accurate movement execution, which we are unaware of, is proprioception, i.e. the sense of body movement and position. The importance of proprioception can be illustrated by the story of Ian Waterman (Rawlence, 1998, *BBC Horizon: The man who lost his body*). At the age of 19 he lost permanently all touch and sense of movement and position below the neck due to, what is believed to be, an auto-immune reaction (McNeill, Quaeghebeur, & Duncan, 2008). When his limbs were out of sight, Ian had no idea where they were. As a result of this lack of all somatosensory feedback of the limbs, the brain could not initiate movement. The immediate behavioural effect was immobility and it was thought that Ian would spend the rest of his life confined to a wheelchair. However, already after a few weeks Ian found out that he was able to move his arms while constantly looking at them. Although the mental effort to do this was enormous, Ian is now able to make movement under visual control. This example is of course highly exceptional. There are only a few people in the world that lost their proprioceptive sense completely, like Ian Waterman. However, an impairment of the proprioceptive sense is not uncommon and can e.g. be seen in children with cerebral palsy (Chrysagis, Skordilis, Koutsouki, & Evans, 2007; Goble, Hurvitz, & Brown, 2009; Wann, 1991; Wingert, Burton, Sinclair, Brunstrom, & Damiano, 2009). Although the motor deficit in SHCP has been examined in great detail, there is still less attention to movement-related sensory impairments, like proprioceptive deficits. Therefore, this thesis will focus on the proprioceptive abilities of the upper limbs in children and adolescents with SHCP and the effects of visual feedback on this ability, i.e. visuo-proprioceptive interactions.

Cerebral Palsy

Cerebral Palsy (CP) is a group of permanent disorders of movement and posture due to a non-progressive lesion in the foetal or infant brain (Bax et al., 2005; Miller, 2007). This lesion can be the result of different factors such as a lack of oxygen to the immature brain, infection, or intoxication (Stanley, Blair, & Alberman, 2000). With an incidence of 2-2.5 per 1000 living births, CP is one of the most common childhood disabilities (Lin, 2003). The classification of CP is typically based on the type of the motor disorder and the number of limbs affected. The former classification encompasses the spastic, dyskinetic, and ataxic form. Ataxia is associated with abnormalities of the cerebellum. It is characterised by loss of orderly muscular coordination. Movements are performed with abnormal force, rhythm and accuracy and low muscle tone is a common feature. About 4% of all CP cases is ataxic. Dyskinetic CP occurs, similar to ataxic CP, especially in term born children. 6% of all CP cases are of the dykinetic subtype. It is the result of lesions to the basal ganglia and is characterised by involuntary, uncontrolled, recurring, occasionally stereotyped movements. The muscle tone is varying and primitive reflex patterns predominate. Finally, the most common subtype is spastic CP, with around 90% of the reported cases. The motor impairment in spastic CP is characterized by an abnormal control of voluntary limb movements, spasticity (i.e. an increased muscle tone and a velocity dependent resistance to stretch which is often related to damage in the motor cortex and/or the pyramidal tract (Dietz & Sinkjaer, 2007; Lance, 1980; Priori, Cogiamanian, & Mrakic-Sposta, 2006), muscle weakness (Ross & Engsberg, 2007), pathological reflexes such as increased reflexes or hyperreflexia and an enduring positive Babinski reflex (indicating a lesion of the pyramidal tract; Krägeloh-Mann & Staudt, 2008). Moreover spastic CP is characterised by an abnormal pattern of movements and posture. In the lower limbs this is visible in equines foot, crouch gait, hip internal rotation and adduction. In the upper limbs this abnormal pattern is characterised by arms in flexion, hands fisted with the thumb adducted or stiff and poorly directed movements of the fingers (Krägeloh-Mann & Staudt, 2008). The more distal body parts are usually affected most. These motor impairments lead to problems with functioning in daily life for walking, reaching and grasping.

In addition to their motor impairments, children with spastic CP also show cognitive problems like learning difficulties, memory deficits, and delayed language development (Bottcher, 2010; Kolk & Talvik, 2000; Krägeloh-Mann & Staudt, 2008). Cerebral visual problems as hemianopsia, blindness and visuo-spatial deficits can also occur in this patient group and epilepsy is commonly seen; it is encountered in about 30%

to 50% of the patients. (Krägeloh-Mann & Staudt, 2008). Moreover, several studies demonstrated that children with SHCP show motor planning deficiencies (e.g. Steenbergen, Meulenbroek, & Rosenbaum, 2004; Steenbergen & van der Kamp, 2004), which may be just as limiting for the performance of activities of daily living as the motor impairments.

Within spastic CP there is a variety of subdivisions[1] (Cans et al., 2007; Krägeloh-Mann & Staudt, 2008). Diplegia/diparesis [2] and quadriplegia/quadriparesis (or tetraplegia/tetraparesis) describe the bilateral involvement, i.e. both sides of the body are affected. In diplegia the legs are more involved than the arms, whereas the term quadriplegia is used only when the arms are as much involved as the legs (diplegia and quadriplegia together account for 60% of all CP cases). In this thesis I will focus on the unilateral spastic subtype of CP, *spastic hemiparetic cerebral palsy (SHCP;* also called spastic hemiplegia). SHCP accounts for 30% of all CP cases and results in motor impairments (see above) that are lateralized to one side of the body (the impaired side of the body, contralateral to the lesioned hemisphere). A lesion on the left side of the brain (left hemispheric lesion; LHL) leads to motor impairments on the right side of the body and a lesion on the right side of the brain (right hemispheric lesion; RHL) results in deficits on the left side of the body. In general, the upper limb is more severely affected than the lower limb. It is therefore not surprising that the manual abilities of the impaired body side in SHCP have been studied extensively. Several studies showed that reaching and grasping with the impaired arm and hand is characterised by an increased movement time, decreased peak velocity, irregular and more segmented movement pattern, and increased trunk involvement. However, a very large variety within and between subjects was reported (Utley & Steenbergen, 2006).

Despite the unilateral character of the disorder, the other side of the body (ipsilateral to the lesioned hemisphere) is not completely free of impairments (less-impaired side of the body; Brown et al., 1989; Gordon, Charles, & Duff, 1999; Steenbergen & Meulenbroek, 2006). Steenbergen and Meulenbroek (2006) for example examined upper limb function for a repetitive reach-and-grasp task towards targets placed at different locations. They showed that movements of the less-impaired side were slower and peak velocity was reached later than in the control group. Moreover, elbow amplitude of this arm was smaller

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[1] In clinical practice, there is currently a tendency to use unilateral CP (i.e. hemiplegia) and bilateral CP (i.e. diplegia and quadriplegia pooled together; Cans, et al., 2007). For reasons of clarity we decided to explain the subtypes as described in different handbooks (Ferrari & Cioni, 2010; Miller, 2007; Stanley, et al., 2000), but note that in literature both terminologies are used.

[2] Literally, ‘plegia’ means complete paralysis whereas ‘paresis’ means partial paralysis or weakening of the muscles. However, in daily practice the terms ‘-plegia’ and ‘-paresis’ are mixed.

for the 60% and 100% arm-length target distances as compared to controls. This is suggested to be due to deficient agonist (Triceps) innervations in the less-impaired arm of the SHCP-group. Despite the deficits of the less-impaired arm, individuals with SHCP usually tend to avoid the use of their impaired arm and are remarkably adept at reaching with the less-impaired extremity towards objects that are located in the contralateral hemispace. In fact, these children actually may have never learned to use their impaired arm for certain motor tasks or may only use it in the simplest manner. The result is that individuals with SHCP tend to perform inherently bimanual tasks of daily living with the less-impaired arm only rather than with both arms (Gordon & Steenbergen, 2008).

Taking into account that a proportion of our daily tasks can be performed with one hand only, the unilateral impairments itself may not largely hamper these children in daily life. Moreover, children with SCHP often develop compensation strategies in order to overcome the unilateral impairment (i.e. they can perform movements with one hand that healthy individuals perform with two hands). Nevertheless, in tasks where the use of both hands is required, the compensations seen in children with SHCP are inefficient and the possible reinforcement of these compensations may make rehabilitation more difficult over time, which highlights the need for early and goal-directed interventions (Charles & Gordon, 2006).

Upper-limb rehabilitation in SHCP

As for any other disorder, rehabilitation of SHCP is a challenge and different approaches to improve the functionality of the impaired arm (sometimes together with the less-impaired arm) do exist, such as constraint-induced movement therapy (CIMT), goal oriented training and bimanual movement therapy. Because each of these approaches is intended to meet a different purpose (Eliasson, 2007), I will not discuss here which approach is the most efficient. For two reasons, I will focus in the remainder of this paragraph on the use of bimanual symmetrical movements in therapy. First, it has been speculated that repetitive training involving symmetrical movements of the impaired and the less-impaired arms might allow the impaired arm to perform at/close to the level of the less-impaired arm. Second, further on in this thesis I will introduce the concept of mirror therapy. This is a specific form of bilateral training and inherently involves bimanual symmetrical movements.

In healthy adults there is a natural tendency towards bimanual symmetry (i.e. inter-limb coupling). The most likely contributors to inter-limb coupling are inter-hemispheric coupling within the cerebral cortex and neural crosstalk. During the performance of bimanual symmetrical movements, simultaneous activation of both hemispheres is often seen and intra-cortical inhibition via the corpus callosum is reduced (Kazennikov et al., 1999; Stinear & Byblow, 2004). Moreover, motor commands generated in the motor cortex are sent to the contralateral side but also to the ipsilateral side of the body (i.e. 10% of the fibers remain uncrossed). This crosstalk is speculated to lead to homologous muscle activation (Cattaert, Semjen, & Summers, 1999). In individuals with unilateral brain damage as in SHCP, bilateral activation does not seem a plausible mechanism to explain the coupling (Volman, Wijnroks, & Vermeer, 2002). Therefore the mechanism of neural crosstalk is believed to play a major role in the coupling between the limbs and the facilitation of the movements of the impaired body side in SHCP.

Indirect support for the use of bimanual symmetrical movements in therapy for SCHP has been provided by studies on the behavioural level (see Goble, 2006 for a review). Sugden and Utley (1995) examined the mutual influence of the impaired and the less-impaired arm in bimanual reaching movements at preferred speed. Temporal synchronization between the hands was found when moving bimanually, but the way in which this was established differed between participants. Either one of the two or both hands adapted during the bimanual movement execution when compared to the unimanual movement. In a follow up study in 1998, Utley and Sugden showed that speeding up the impaired hand resulted in a stronger coupling between the hands, particularly in the first part of the movement (Utley & Sugden, 1998). Steenbergen, Hulstijn, de Vries and Berger (1996) showed, in a separate series of studies, similar coupling between the hands when participants were asked to place as quickly as possible two balls in a hole (one with each hand). However, in this study the temporal coupling was established in a uniform manner, i.e. for all participants the less-impaired arm slowed down under bimanual responding whereas the performance of the impaired arm was relatively unaffected. In addition, Volman, Wijnroks and Vermeer (2002) showed that bimanual symmetrical movements may facilitate and enhance the movement of the impaired arm in SHCP. They compared unimanual and bimanual performance for a circle drawing task. In the unimanual condition, performance of the impaired arm was less smooth and more variable than that of the less-impaired arm. However, moving both arms in a symmetric fashion resulted for the impaired arm in smoother and less-variable movements when compared to the unimanual condition.

Taken together, these results suggest that despite their unilateral impairment, individuals with SHCP are able to couple their movements to a similar extent as typically developing (TD) people. In performing these bimanual symmetrical movements, the less-impaired arm might be useful in providing a template for the impaired arm and this might enhance impaired upper limb performance (within a single session). However, until now studies have mainly focused on kinematic variables (such as speed, trajectory or timing of the two limbs) and it remains to be determined whether bimanual symmetry has an effect on proprioception as well.

Proprioception

When we close our eyes we still know where our body parts are in space and relative to each other. This sense is termed proprioception and consists of two components: (joint-) position sense (the sense of static limb position) and kinaesthesia (the sense of limb movement). Proprioception is mediated by so called propioceptors in the skin, muscles, tendons, ligaments and joint capsules (Proske & Gandevia, 2009; Sherrington, 1906). The receptors in the muscles, the muscle spindles, are accepted to make a major contribution to proprioception. The primary endings of the muscle spindle respond to changes in the size of the muscle length and its speed and are therefore believed to contribute to both position sense and kinaesthesia. The secondary endings of the muscle spindle signal the change of the length and therefore only contribute to the sense of position (Proske & Gandevia, 2009; Sherrington, 1906).

Proprioception is essential for movement performance and has been shown to be important in the production of coordinated movements in multiple ways (Goble, Lewis, Hurvitz, & Brown, 2005). It plays a major role in controlling muscle interaction torques (Sainburg, Ghilardi, Poizner, & Ghez, 1995), in timing the coordination between limb segments (Cordo, Carlton, Bevan, Carlton, & Kerr, 1994), in monitoring movement trajectories (Ghez, Gordon, Ghilardi, Christakos, & Cooper, 1990), and in establishing internal representations used during the acquisition and adaptation of skilled movement (Kawato & Wolpert, 1998). It is therefore not surprising that impaired proprioception is found to be implicated in motor disorders such as hemiparetic stroke (Niessen et al., 2008) or CP (e.g. Chrysagis et al., 2007; Cooper, Majnemer, Rosenblatt, & Birnbaum, 1995; Opila-Lehman, Short, & Trombly, 1985; Wingert et al., 2009).

Research has shown that during motor development and learning, a shift in reliance from vision to proprioception takes place (Fleishman & Rich, 1963; Smyth & Marriott, 1982). It is suggested that monitoring of limb movements is delegated from vision to proprioception as learning proceeds (Smyth & Marriott, 1982). Moreover, Fleishman and Rich (1963) showed that individuals with high proprioceptive sensitivity (measured as small difference limens for judgments of lifted weights) could make use of this proprioceptive information during a practice period of a two-hand coordination task and were suggested to be able to switch rapidly from a visual to a proprioceptive control of movement. In contrast, individuals who relied more on visual information made a rapid progress in the beginning of learning but could not switch as accurate as the other group from visual to proprioceptive control during learning. In individuals with SHCP both learning and the shift from vision to proprioception during learning are thought to be considerably hampered due to a disturbed proprioception of the impaired arm (Chrysagis et al., 2007; Goble et al., 2009; Wingert et al., 2009) and an increased reliance on visual information (Verrel, Bekkering, & Steenbergen, 2008). Therefore, any therapeutic intervention that aims to improve motor function with the involvement of visual feedback in children with SHCP depends on its effect on proprioception.

Different studies already examined proprioception in SHCP and showed predominantly deficits of the impaired arm (Chrysagis et al., 2007; Goble et al., 2009; Wingert et al., 2009). However, proprioception in itself is difficult to evaluate because different factors, such as memory, can affect the measurement. The studies described in this thesis focused on one aspect of proprioception, i.e. the sense of static limb position or position sense. Different methods to measure position sense are reported in literature (Goble et al., 2005) and have been used in the examination of proprioception in SHCP. In ipsilateral matching tasks the same arm serves both as reference arm and as matching arm. It is thus inherent to the task that participants need to memorize the target position to match it accurately. Children with CP are prone to having memory problems (Bottcher, 2010) and thus it is likely that a portion of the matching error reflects cognitive and/or memory deficits rather than a deterioration of proprioception (Goble, 2010). A similar problem occurs for the contralateral remembered matching task in which one (reference) hand is moved to the target and (after a few seconds) is returned to the start position. Subsequently, the participant is required to reproduce the same movement with the contralateral hand. To circumvent the involvement of memory in this thesis we used a contralateral matching task to measure position sense (chapter three) and visuo-proprioceptive interactions (chapter four). In this task, the reference arm is moved to a target and remains there while the participant matches this target location with the contralateral hand. There is thus ‘online’ proprioceptive information about the reference position available. With this task it is difficult to pinpoint whether the error that is measured arises from one arm or the other (Goble, 2010), but it can provide information about how problems with proprioception influence tasks that involve both arms. This is particular relevant for the study of children with SHCP whose motor impairments are lateralized to one body side but are known to hamper bimanual actions.

The validity and reliability of position matching tests have rarely been evaluated, but it is generally accepted that the magnitude of the matching errors is a useful indicator of position sense (Goble, 2010). In this thesis it was therefore chosen to take the *absolute matching error* as a measure for the *matching accuracy*, which in turn is an indicator of position sense. The absolute matching error is the absolute difference in centimetres at the end of the movement between the moving hand and the target position (defined by the contralateral hand [chapter three and four] or by an external visual target [chapter five and six]). A shorter distance/smaller error is related to a higher movement accuracy and thus indicates a better position sense.

Mirror visual feedback

Mirror visual feedback is created by placing a mirror in between the two upper limbs along the mid-sagittal plane. The reflection of one limb seen in the mirror is superimposed on the position of the limb behind the mirror (Altschuler et al., 1999; Holmes & Spence, 2005; Ramachandran & Rogers-Ramachandran, 1996). When now moving the limbs, the illusion is created of a zero lag symmetric movement between the two arms (Altschuler et al., 1999; Ramachandran & Rogers-Ramachandran, 1996). The use of mirror visual feedback in experimental studies is twofold: on the one hand it is used to manipulate visual feedback to create a conflict between the visual and proprioceptive information (e.g. by visually manipulating the position of a hand before the start of a movement). In doing so one can examine e.g. the relative ‘weighting’ of two sources of sensory information (i.e. vision and proprioception; Holmes & Spence, 2005). On the other hand, studies examined the effects of mirror visual feedback on movement performance in patients with unilateral pain and movement disorders to get more insight into its possible application in therapy (e.g. Altschuler et al., 1999; McCabe et al., 2003).

Ramachandran and Rogers-Ramachandran (1996) were the first to describe the use of mirror visual feedback in the treatment of phantom limb pain in amputees. After a short period of ‘mirror therapy’, which involved bilateral mirror-symmetric movements, amputees reported a decrease in phantom pain. Based on the effect of visual feedback through a mirror in patients with phantom limb pain, a number of subsequent studies were performed on the effects of mirror visual feedback in other acquired unilateral motor or pain disorders. It was found that chronic stroke patients could benefit from this type of therapy, showing increases in range of motion, speed and accuracy of arm movements

(Altschuler et al., 1999; Stevens & Stoykov, 2003), an improved functional use and a recovery of grip strength (Sathian, Greenspan, & Wolf, 2000). Likewise, in patients with Chronic Regional Pain Syndrome 1 (CRPS1) mirror visual feedback of the unaffected limb reduced the perception of pain and stiffness (McCabe et al., 2003).

Mirror visual feedback is suggested to act by restoring the congruence between motor output and sensory input (Ramachandran, 2005; Ramachandran & Altschuler, 2009). In individuals without movement impairment, motor commands sent from the motor cortex are normally damped by sensory feedback. However, if a movement is impaired there is a discrepancy between the centrally generated efference copy of the motor commands and the sensory feedback. This is thought to amplify the motor output, which in turn is suggested to deteriorate motor performance even further. Mirror visual feedback may act by interrupting this ‘loop’. In other words, the mirror provides patients with ‘proper’ visual input which is suggested to reduce movement difficulties and reverse elements of learned disuse of the impaired arm which in turn could lead to a ‘relearning’ of the use of the impaired arm (Altschuler et al., 1999).

In addition to the studies on mirror visual feedback in acquired disorders like stroke and CRPS1, more recent studies examined the effects of mirror visual feedback in a patient group with a congenital unilateral disorder. Feltham, Ledebt, Bennett, Deconinck, Verheul, and Savelsbergh (2010) recently showed that the positive effects of mirror visual feedback may potentially be extended to individuals with congenital disorders such as SHCP. When performing a symmetrical bimanual circular movement, mirror visual feedback reduced the movement variability in comparison with a condition in which only the less-impaired limb was visible. Moreover, mirror feedback resulted in a reduction of the excessive neuromuscular intensity in the shoulder muscles and a decrease in undue eccentric and concentric activity in the elbow muscles of the impaired limb, indicating improved efficiency (Feltham, Ledebt, Deconinck, & Savelsbergh, 2010). According to Feltham, Ledebt, Deconinck et al. (2010) and Feltham, Ledebt, Bennett et al. (2010) these results suggest that mirror visual feedback can be used to improve the motor control in children with SHCP and could thus be suitable for non-acquired disorders as well. Indeed, a more recently published study by Gygax, Schneider and Newman (2011) showed improvements in grip strength and the position of the upper limb during achievement of specific tasks (dynamic position analysis; subscale of the SHUEE assessment). To summarize, mirror visual feedback seems to have a positive effect on different aspects of movement in individuals with SHCP such as the excessive eccentric muscle activity, force and movement symmetry. However, literature on this topic is still scarce and more research is needed in order to scrutinize the effects of mirror visual feedback on other factors that are essential for movement performance such as proprioception.

Outline of the thesis

The work presented in this thesis followed on previous work of Feltham, Ledebt, Deconinck et al. (2010) and Feltham, Ledebt, Bennett et al. (2010) who were the first to examine the effects of mirror visual feedback on movement behaviour and neuromuscular activity in children with SHCP. Despite the fact that they reported positive effects of mirror visual feedback in SHCP, it remained unclear from their studies whether the positive effects were the result of visual symmetry (irrespective of which arm is viewed in the mirror) or of the illusion that the impaired arm has been substituted by the mirror image of the less-impaired arm. Our first study was designed to answer this question. As described in **chapter two**, we compared two situations on the level of movement kinematics and neuromuscular activity: the mirror condition and the reversed mirror condition. In the mirror condition participants received mirror visual feedback of their less-impaired arm whereas in the reversed mirror condition participants received mirror visual feedback of the impaired arm. By this means we could get more insight into the positive effects of mirror visual feedback in SHCP as reported by Feltham, Ledebt, Deconinck et al. (2010) and Feltham, Ledebt, Bennett et al. (2010). Subsequently, we were interested in the effects of mirror visual feedback on one aspect of proprioception, position sense. In **chapter three** we therefore first measured position sense with a contralateral matching task. This task can provide us with important information about how problems with proprioception may affect movements that involve both arms. One arm was fixed on a target position and participants were asked to match the other arm into the same (mirror symmetric) position while no visual information of either arm was available. In order to get good insight into their deficiencies we compared the SHCP children with typicalle developing (TD) peers. In **chapter four** we then scrutinized the effects of (mirror) visual feedback of a static reference arm on the position sense of the moving matching arm in individuals with SHCP. Previous studies in TD children showed that visual information of a static reference hand improved matching accuracy (Von Hofsten & Rösblad, 1988), but for individuals with quadriplegia with bilateral brain damage no such improvement was found (Wann, 1991). Individuals with SHCP have unilateral brain damage and thus we were interested whether visual feedback of a reference arm could improve the matching accuracy of the matching arm in this patient group. A similar contralateral matching task as in chapter three was used, but now an opaque screen or a mirror was placed in between the arms in the sagittal plane so that the impaired arm was invisible. In the screen condition the participants could see their less-impaired arm, in the mirror condition the less-impaired arm and its mirror reflection was visible. These two conditions were compared with a condition in which the participants did not receive any visual feedback of their movement.

In chapters five and six we aimed to get more insight into the possibilities to use mirror visual feedback in the rehabilitation of individuals with SHCP. In **chapter five** the aim was twofold: on the one hand we aimed to examine the effect of bimanual symmetrical movements on matching accuracy. To this end, we compared for the impaired arm the accuracy of matching a visual target under unimanual and bimanual conditions. On the other hand we aimed to examine the effects of mirror visual feedback during bimanual symmetrical movements on the matching accuracy of the impaired arm. We placed an opaque screen or a mirror in between the arms in the sagittal plane so that participants either saw their less-impaired arm (screen) or their less-impaired arm and its mirror reflection (mirror). Matching accuracy in these two conditions were compared to reveal the effects of mirror visual feedback. The studies reported in chapter three, four and five all examined 'immediate' effects of (mirror) visual feedback, while the effects of mirror visual feedback after a short period of practice remained to be determined. Therefore, **chapter six** describes the effects of a short practice of a matching movement with (mirror) visual feedback. Children and adolescents with SHCP performed a 20 minute bimanual practice with mirror visual feedback (mirror-group) or 'regular' visual feedback of the less-impaired arm (screen-group). In the pre-, post-, and retention-test the matching accuracy was determined (without visual feedback) and compared between the two training groups. The general discussion in **chapter seven** summarizes the findings of each chapter and discusses the main results. At last, suggestions for future research are given.

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Chapter 2

The mirror reversed

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Smorenburg, A.R.P., Ledebt, A., Feltham, M.G., Deconinck, F.J.A., Savelsbergh, G.J.P. (2011). The positive effect of mirror visual feedback on arm control in children with Spastic Hemiparetic Cerebral Palsy is dependent on which arm is viewed. *Exp Brain Res, 213(4):* 393-402.

Abstract

Mirror visual feedback has previously been found to reduce disproportionate interlimb variability and neuromuscular activity in the arm muscles in children with Spastic Hemiparetic Cerebral Palsy (SHCP). The aim of the current study was to determine whether these positive effects are generated by the mirror per se (i.e. the illusory perception of two symmetrically moving limbs, irrespective of which arm generates the mirror visual feedback) or by the visual illusion that the impaired arm has been substituted and appears to move with less jerk and in synchrony with the less-impaired arm (i.e. by mirror visual feedback of the less-impaired arm only). Therefore we compared the effect of mirror visual feedback from the impaired and the less-impaired upper limb on the bimanual coupling and neuromuscular activity during a bimanual coordination task. Children with SHCP were asked to perform a bimanual symmetrical circular movement in three different visual feedback conditions (i.e. viewing the two arms, viewing only one arm, and viewing one arm and its mirror image), combined with two head orientation conditions (i.e. looking from the impaired and looking from the less-impaired body side). It was found that mirror visual feedback resulted in a reduction of the eccentric activity of the Biceps Brachii Brevis in the impaired limb compared to the condition with actual visual feedback from the two arms. More specifically, this effect was exclusive to mirror visual feedback from the less-impaired arm and absent when mirror visual feedback from the impaired arm was provided. Across conditions the less-impaired arm was the leading limb, and the nature of this coupling was independent from visual condition or head orientation. Also, mirror visual feedback did not affect the intensity of mean neuromuscular activity or the muscle activity of the Triceps Brachii Longus. It was concluded that the positive effects of mirror visual feedback in children with SHCP are not just the result of the perception of two symmetrically moving limbs. Instead, in order to induce a decrease in eccentric neuromuscular activity in the impaired limb, mirror visual feedback from the ‘unaffected’ less-impaired limb is required.

Introduction

Children with Spastic Hemiparetic Cerebral Palsy (SHCP), who have unilateral motor impairments in both their arm and leg due to brain and/or pyramidal tract damage (Miller, 2007) [1], perform tasks requiring only the less-impaired hand reasonably well (e.g. Steenbergen, Hulstijn, de Vries, & Berger, 1996; Utley & Sugden, 1998). In contrast, tasks requiring bimanual coordination pose a huge challenge because of the inevitable involvement of the impaired arm and hand. In recent years, bimanual reaching and grasping has been thoroughly investigated in individuals with SHCP (e.g. Steenbergen et al., 1996; Sugden & Utley, 1995; Utley & Sugden, 1998; Volman, Wijnroks, & Vermeer, 2002). Interestingly, these studies suggest that, despite the unilateral impairment, bimanual actions of children with SHCP seem to be facilitated by bilateral connections at multiple levels of the central nervous system similar to what has been found in typical populations (e.g. corticospinal, cerebellar, brain stem, and propriospinal; Wiesendanger, Kaluzny, Kazennikov, Palmeri, & Perrig, 1994). For example, Volman et al. (2002) showed that when drawing circles in an in-phase (symmetrical) coordination mode the spatiotemporal interlimb variability decreased. Furthermore, movement smoothness of the impaired limb increased compared to single-handed performance. Steenbergen, Charles and Gordon (2008) observed close temporal synchrony of the hands when grasping an object bimanually, which contrasted with the timing differences between both hands when they performed separately. It should be noted that some of these findings indicate adaptations of the less-impaired side to the behaviour of the affected side (e.g. Steenbergen et al., 1996), but combined these studies suggest that bilateral interactions exist in children with SHCP and that they can lead to favourable effects in the impaired arm.

A paradigm that has been used to further our understanding of how visual and spatial processes influence coordination and perception of the two hands is the ‘mirror box illusion’ (e.g. Franz & Packman, 2004; Holmes & Spence, 2005). This illusion is manifested when a mirror is placed in between the two upper limbs along the mid-sagittal plane. The reflection of the arm viewed in the mirror seems superimposed on the visual image of the arm behind the mirror. When the arm facing the reflective side is moved this creates the illusory perception of a zero lag symmetrical movement of the two limbs. The effects of mirror visual feedback were first investigated by Ramachandran and Rogers-

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[1] Cerebral Palsy (CP) is a group of permanent disorders of movement and posture due to a non-progressive lesion in the fetal or infant brain (Miller, 2007). CP is the most common cause of childhood disability and has an incidence of 2-2.5 per 1000 living births (Lin, 2003). A common form of CP is Spastic Hemiparetic Cerebral Palsy (SHCP). Children with SHCP have a brain lesion in one hemisphere and as a result have spasticity on the other side of the body.

Ramanchandran (1996) in amputees with phantom pain. After a short period of 'mirror box' therapy, which involved (bilateral) mirror-symmetric movements, amputees reported a decrease in phantom pain. These encouraging findings led to the adoption of mirror visual feedback in treating other acquired unilateral motor or pain disorders where the illusion appeared to result in positive effects on motor performance and pain perception (for a review see Ramachandran & Altschuler, 2009). For instance, it was found that chronic stroke patients could benefit from therapy using mirror visual feedback, showing increases in range of motion, speed and accuracy of arm movements (Altschuler et al., 1999; Stevens & Stoykov, 2003), and an improved functional use and a recovery of grip strength (Sathian, Greenspan, & Wolf, 2000). Likewise, in patients with Chronic Regional Pain Syndrome 1 (CRPS1) mirror visual feedback of the unaffected limb reduced the perception of pain and stiffness (McCabe et al., 2003).

Interestingly, Feltham, Ledebt, Bennett et al. (2010) and Feltham, Ledebt, Deconinck et al. (2010b) demonstrated that the positive effects of mirror visual feedback may potentially be extended to individuals with congenital disorders such as SHCP, a finding that was recently supported by Gygax, Schneider and Newman (2011) who showed that mirror therapy in children with hemiplegia may improve strength and dynamic function of the impaired arm. Feltham, Ledebt, Bennett et al. (2010) and Feltham, Ledebt, Deconinck et al. (2010b) used a task where participants performed continuous symmetrical circular movements with both upper limbs in three visual conditions (glass: seeing the two arms; screen: seeing only the less-impaired arm; mirror: seeing the less-impaired arm and its mirror reflection). An effect of mirror visual feedback was found on the nature of the bimanual coordination (Feltham, Ledebt, Bennett et al., 2010) and on the neuromuscular activation in children with SHCP (Feltham, Ledebt, Deconinck et al., 2010b). More specifically, in the first study it was demonstrated that movement variability of the interlimb coupling was lower in the mirror condition in comparison with the screen condition. In addition, mirror visual feedback resulted in a reduction of the neuromuscular intensity in the shoulder muscles of the less-impaired limb and a shortening of the duration of eccentric and concentric activity in the elbow muscles of the impaired limb. In accordance with Perry, Davis and Luciano (2001), a phase where a flexor muscle (e.g. Biceps Brachii Brevis, BBB) was actively contributing to a flexion movement was defined as *concentric*, whereas flexor activity was *eccentric* when it contributed to an extension movement. For extensor muscles (e.g. Triceps Brachii Longus, TBL) the opposite classification was used. Note that an earlier study showed that children with SCHP performed this bimanual coordination task with higher levels of neuromuscular intensity in elbow and wrist muscles and longer periods of concentric and eccentric activity in elbow and shoulder muscles compared to typically developing children (Feltham, Ledebt, Deconinck, & Savelsbergh, 2010a). More eccentric activity of the BBB might suggest more counteraction to the extension movement, and hence indicates that the neuromuscular control is less efficient in children with SHCP. The finding of a decrease in interlimb variability and a reduction of eccentric and concentric muscle activity in a condition with mirror visual feedback thus shows that the mirror has the capacity to induce a general improvement of the kinematics and the neuromuscular efficiency during bimanual movements in children with SHCP.

A pertinent question is, however, whether the mirror effects observed in these children are caused by the illusory perception of seeing two arms moving in perfect symmetry, irrespective of which arm is seen in the mirror, or by the illusion that the impaired limb has been substituted with a less-impaired limb, which is not spastic. The studies by Feltham, Ledebt, Bennett et al. (2010) and Feltham, Ledebt, Deconinck et al. (2010b) described above have only investigated the effect of mirror visual feedback from the unaffected arm and therefore were not able to discriminate between these two explanations. When Franz and Packman (2004) found that mirror visual feedback was powerful enough to enhance spatial coupling of the two hands in healthy adults performing a circle drawing task in a similar manner as actual vision of both hands, this effect was independent of the laterality of the mirror visual feedback. In a condition where only one hand was visible, the circles drawn by the hand in vision were found to be significantly larger than for the hand hidden behind the screen. Mirror visual feedback, regardless of which hand was viewed, had the capacity to wipe out this between-hand difference in circle size. Franz and Packman (2004) hypothesised that the illusion of the perfect symmetry between the two hands created by the mirror promoted the sensorimotor coupling at the central level.

In children with SHCP, however, the movement produced by the impaired and less-impaired arm is qualitatively different, and hence the mirror visual feedback created by either arm is considerably different as well. Whilst there is an illusion of perfect symmetric movement in both situations, the mirror visual feedback of the impaired arm shows a less smooth movement hampered by the motor deficits. This discrepancy between the two sides and the mirror visual feedback they elicit enables us to investigate the mirror box illusion in this group of children in more detail. More specifically, the aim of the present study was to determine whether the mirror effects as found previously by Feltham, Ledebt, Bennett et al. (2010) and Feltham, Ledebt, Deconinck et al. (2010b) are the result of the perception of visual symmetry per se, irrespective of which arm is viewed, or by the illusion that the impaired arm has been substituted and appears to move smoother and in synchrony with the less-impaired arm. For this purpose, we compared the effect of mirror visual feedback generated by the less-impaired and the impaired arm on the bimanual coupling and the neuromuscular activity in children with SHCP during a bimanual coordination task similar to the one used by Feltham, Ledebt, Bennett et al. (2010) and Feltham, Ledebt, Deconinck et al. (2010b). Based on the studies of Feltham and colleagues we anticipate that mirror visual feedback from the less-impaired arm will result in increased interlimb coupling and reduced eccentric activity in the arm muscles of the impaired limb compared to the visual feedback of both arms (glass condition). If the illusion of visual symmetry is the main trigger for the changes induced by the mirror, mirror visual feedback of the less-impaired arm is expected to induce similar effects on the kinematics and the neuromuscular activity as compared to mirror visual feedback of the impaired arm. Alternatively, if the mirror effect in children with SHCP is caused by a mechanism involving substitution of the visual information of the impaired arm by visual feedback from the less-impaired arm, we expect to find less favourable changes to the control of the movement when viewing the impaired upper limb and its mirror reflection than when viewing mirror visual feedback of the less-impaired limb.

Methods

Participants

Ten children (eight males and two females) with SHCP participated in the study (mean age 12.7 ± 3.2 years). Further participant characteristics can be found in Table 2.1. A subset of the data from seven children who took part in a previous study (Feltham, Ledebt, Deconinck et al., 2010b) was identified to be included in the present analysis. The participants did not have impaired vision or any neuromuscular disorders other than SHCP. Severity of the impairment was assessed by a single experimenter with the Modified Ashworth Scale (MAS; spasticity levels increase from 1 to 4), Gross Motor Function Classification System (GMFCS; function deteriorates from I to V) and the functional independence measure for children (WeeFIM; motor items only, with a possible score range of 13 to 91. A higher score denotes a better functional independence of the child). Written informed consent was obtained from all participating children and their parents. The experiment was conducted in accordance with the Declaration of Helsinki and all experimental procedures were approved by the institutional research ethics committee.

Table 2.1: Participant characteristics. For each participant the age in years, sex, impaired arm, MAS, GMFCS and WeeFIM score and aetiology are represented.

P	Age (years)	Sex	Impaired arm	MAS	GMFCS	WeeFIM	Aetiology
1	12.8	M	Right	1	I	90	Unknown
2	9.3	F	Right	1+	I	89	Cerebral haemorrhage
3	13.2	M	Right	1	I	91	Unknown
4	14.3	M	Right	1+	I	91	Cerebral haemorrhage during birth and meningitis just after birth
5	11.0	M	Right	1	II	55	Meningitis just after birth
6	6.8	M	Right	1	I	83	O2 shortage during birth
7	17.1	M	Right	2	I	91	Cerebral haemorrhage
8	11.1	M	Left	1	I	91	Unknown
9	14.7	M	Left	2	II	62	Schizencephaly
10	16.3	F	Left	1	I	79	O2 shortage during birth

Test procedures

Each participant was seated on a height adjustable chair at a table with both feet flat on the floor and the knees 90° flexed. The elbows were flexed over 90° and in each hand the participant grasped a handle attached to a wooden disc (radius 0.10 m) which spun freely 360° around a vertical axis. The axes were fixed to a wooden plateau and were located 0.31 m apart.

Participants were asked to perform a continuous inward symmetrical circular bimanual movement (the right arm rotated anti-clockwise and the left arm rotated clockwise). Starting at the inner most part of each circle (nine o'clock for the right arm and three o'clock for the left arm) children were asked to rotate the discs continuously at a self-selected speed until they were instructed to stop. Additionally, they were instructed to keep the movement time per cycle (i.e. movement frequency) constant across the experimental trials and the different conditions. The type of visual feedback was varied so that the participant 1) viewed both arms, 2) viewed only one arm, 3) viewed one arm and its mirror reflection, by placing a glass, opaque screen, or mirror divide, respectively (all: width 0.06 m, depth 0.75 m, height 0.39 m), between the arms along the mid-sagittal plane (Figure 2.1). The glass and the screen condition were added as control conditions. In addition, in order to examine the difference between mirror visual feedback of the less-impaired arm (referred to as 'uncompromised' mirror visual feedback) and mirror visual feedback of the impaired arm (referred to as 'compromised' mirror visual feedback) on the nature of the bimanual coupling and the neuromuscular activity in the BBB and TBL muscle the orientation of the head (i.e. viewing side) was varied; the participants orientated their head either towards the impaired side of the body (ViewImp) or towards the less-impaired side of the body (ViewLessImp).

The six conditions (3 visual feedback x 2 viewing side conditions) were presented in a random order and per condition, three trials, each lasting approximately 15 seconds, were recorded. Prior to data collection, practice trials were conducted to familiarise the participants with the test setup. Short breaks were given between the trials in order to recover from any fatigue or decrease in concentration that might have occurred during the performance of the experiment. In order to keep the participants motivated they were told that rotating the discs more symmetrically resulted in more points. At the end of the experiment the children could trade their points for a small gift.

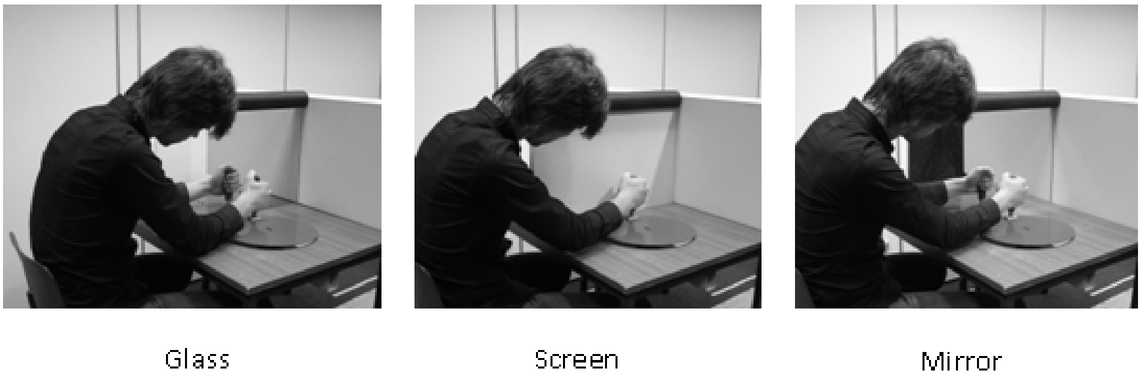

Figure 2.1: Experimental setup showing one of the experimenters demonstrating the task during the glass (left panel), screen (middle panel), and mirror (right panel) condition. The participant viewed the bimanual task either from the impaired or from the less-impaired side of the body. Note that the participants were considerably smaller than the experimenter and that their posture was more erect than shown in this picture.

Recording and analysis procedures

The 3D position of the wrist, elbow and shoulder was determined by two serially-connected units containing three infrared cameras at 200 Hz (3020 Optotrak, Northern Digital Inc., Waterloo, Canada). Light emitting diodes were bilaterally attached to the skin with double-sided tape over the dorsal tuberculum of the radius (wrist), lateral epicondyle of the humerus (elbow), greater tubercle of the humerus (shoulder) and the trochantor of the femur (hip). The phase of each limb was calculated according to the following formulas:

$\varphi\mathrm{D} = \arctan\,[(\mathrm{d}S_\mathrm{D} \cdot \mathrm{d}t^{-1}) \,/\, S_\mathrm{D}]$,

and

$\varphi\mathrm{ND} = \arctan\,[(\mathrm{d}S_\mathrm{ND} \cdot \mathrm{d}t^{-1}) \,/\, S_\mathrm{ND}]$,

where $\varphi\mathrm{D}$ and $\varphi\mathrm{ND}$ are the phase of the dominant (less-impaired) and the non-dominant (impaired) hand respectively, S_D and S_ND are the position time series, and $\mathrm{d}S_\mathrm{D} \cdot \mathrm{d}t^{-1}$ and $\mathrm{d}S_\mathrm{ND} \cdot \mathrm{d}t^{-1}$ represent the instantaneous velocity. Before the calculation of $\varphi\mathrm{ND}$, the sign of the position time series of the non-dominant arm was inversed to an anti-clockwise trajectory. The continuous relative phase (CRP) indicating the degree of coupling (i.e. synchronicity) between the arms is then:

$CRP = \varphi\mathrm{D} - \varphi\mathrm{ND}$,

where a positive value for CRP implied the less-impaired arm lead and a negative value the impaired arm lead.

Superficial EMG (electromyography) was bilaterally recorded from the main muscles around the elbow: the Biceps Brachii Brevis (BBB) and the Triceps Brachii Longus (TBL), according to the SENIAM guidelines for surface EMG measurement (Hermens, Freriks, Disselhorst-Klug, & Rau, 2000). The ground electrode was placed over the acromion on the side of the less-impaired hand. Disposable Ag/AgCl surface EMG electrodes with a gel-skin contact, active detection area of 15mm^2 for each electrode and a 20mm centre to centre inter-electrode distance, were placed in parallel with the muscle fibre direction over the muscle bellies after cleaning and gentle abrasion of the skin. The EMG signals were amplified 20 times high-pass pre-filtered at 10 Hz and AD-converted at 1000 Hz with a 22-bit resolution and stored on a computer. The EMG signals were band-pass filtered with a zero lag 2nd order Butterworth filter between 10 and 400 Hz and then full-wave rectified. Finally, the EMG signals were smoothed with a zero-lag 2nd order low-pass Butterworth filter at 6 Hz.

Bilateral EMG recordings were analyzed from the first two cycles of each trial[2]. Typically, EMG amplitudes are scaled to activation levels recorded either during an isometric maximal voluntary contraction or a specified steady-state sub-maximal contraction. However, this procedure is likely to be unreliable in people with neurological conditions since they are often unable or unwilling to perform maximum contractions (Smith, Coppieters, & Hodges, 2008; van Dieën, Selen, & Cholewicki, 2003). Therefore, to determine the intensity of the mean neuromuscular activity of each muscle during the bimanual movement, the mean amplitude was calculated from the smoothed raw EMG signals. In addition, the amount of concentric and eccentric muscle activity was determined. To this end, the EMG profile of each muscle was broken down into active and inactive phases, after the threshold for muscle contraction was determined. Consistent with Perry et al. (2001) it was assumed that a purposeful activation of a muscle causes an increase in the EMG signal within the frequency range of 0 – 160 Hz. The active/inactive threshold value was then calculated as follows: $T = 15 + 1.5R$, where T is the threshold value, R is the mean value of the EMG signal above 160 Hz and the constants are derived from Perry et al. (2001). A muscle was classified as active if the smoothed raw EMG signal was above the threshold level. Subsequently, the active phases were classified as eccentric, concentric, or isometric depending on the observed elbow movement and the primary mechanical function of the muscle (i.e. flexion or extension). For example, BBB muscle activity above threshold was classified as concentric when the elbow was being flexed and as eccentric when the elbow was being extended. Above threshold TBL muscle activity was classified as concentric for elbow extension and as eccentric activity for elbow flexion. If the muscle was active but no change in elbow angle was observed, it was classified as isometric activity. However, this isometric activity was not included in further analysis of this study since the task involved a dynamical movement with accordingly very short relative durations of isometric activity (1.25% of the total muscle activity). The duration of all eccentric and concentric phases was summed and expressed as a percentage of the total movement time (i.e. the movement time of the first two cycles), giving the relative duration of eccentric activity and the relative duration of concentric activity for each muscle.

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[2] Only the first two cycles of each trial could be analyzed since some children with SHCP could only fulfil 2 cycles before they adopted a different coordination mode than the one they were instructed to produce. Moreover, for some children the movement time allowed them to complete only 2 cycles within the allocated time of each trial or the hand slipped off the handle at which point the trial had to be terminated.

Statistical analysis

The effect of viewing side and visual feedback condition on the bimanual coupling, EMG intensity and the phases of muscle activity in each arm, was tested using a repeated measurement ANOVA with three within factors: Limb (impaired, less-impaired), Viewing side (view impaired [ViewImp], view less-impaired [ViewLessImp]), and Visual condition (mirror, screen, glass). These analyses were conducted using mean data calculated from the three trials per combination of independent variables. In the event that the sphericity assumption was violated, Greenhouse-Geisser adjustments were applied. Fisher's LSD tests were used for post-hoc analysis and the level of significance was set at 0.05.

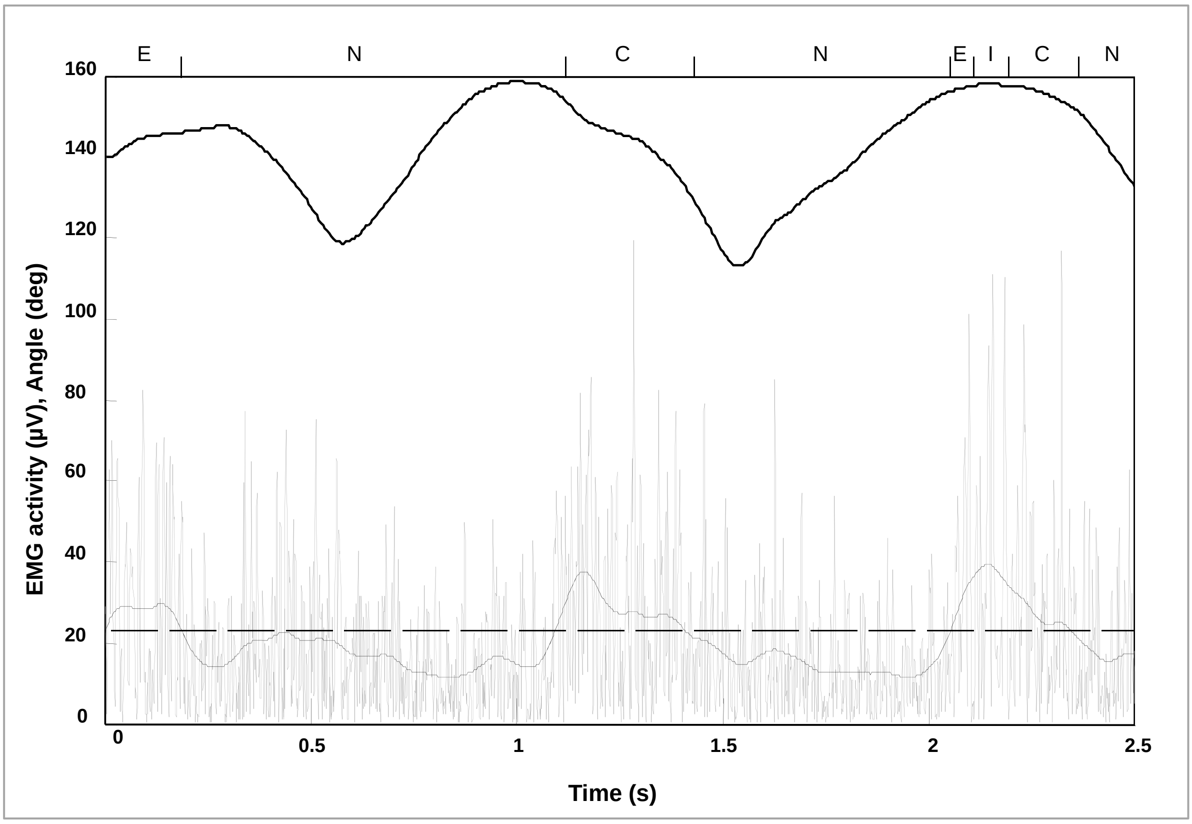

Figure 2.2: Data from a representative trial showing the rectified EMG activity (light grey) from the Biceps Brachii Brevis and the smoothed EMG (dark grey line). In addition, the elbow angle (thick black line) and the active/inactive threshold for the muscle contraction (dashed line) are depicted. Muscle activation is classified as eccentric (E), concentric (C), isometric (I) and inactive (N).

Results

Bimanual coupling

The CRP did not significantly differ between the three visual conditions (mirror = 6.6° ± 6.3°; screen = 13.2° ± 7.2°; glass = 10.8° ± 7.4°) and the viewing side did not have an effect on the interlimb coupling either (ViewImp = 11.1° ± 6.4° and ViewLessImp = 9.3° ± 7.0°; see Table 2.2 for values per individual condition). The overall mean was 10.2° ± 6.6°, indicating that the less-impaired arm was the leading limb.

Table 2.2: Mean and SE values of the continuous relative phase (CRP) in degrees for each visual condition and viewing condition.

	ViewImp	ViewLessImp
Mirror	8.1 ± 7.7	5.0 ± 6.6
Screen	17.2 ± 7.1	9.3 ± 8.6
Glass	8.0 ± 6.6	13.6 ± 8.6

Intensity of the mean neuromuscular activity in BBB and TBL

There were no significant main or interaction effects on the mean neuromuscular activity in BBB and TBL of either Viewing side or Visual condition (see Table 2.3). This means that the mean EMG intensity in BBB and TBL did not change as a function of viewing side or the nature of visual feedback. Viewing the impaired arm and its mirror reflection did not result in higher levels of EMG intensity (BBB: 24.1 ± 3.1 μV; TBL: 9.9 ± 1.2 μV) than viewing the less-impaired arm and its mirror reflection (BBB: 21.7 ± 3.6 μV; TBL: 11.2 ± 2.0 μV). Inspection of Table 2.3 seems to indicate a trend ($F_{2,18} = 2.76$, $p = 0.09$) towards lower intensities of neuromuscular activity in the mirror condition compared to the glass and screen condition (especially in the BBB of the less-impaired limb in the ViewLessImp condition). In addition, the mean neuromuscular intensity tended to be higher in the impaired than in the less-impaired arm for both the BBB and TBL muscles (BBB: 29.0 ± 4.9 μV vs. 19.5 ± 3.9 μV; TBL: 14.7 ± 3.3 μV vs. 8.5 ± 1.1 μV), however, the ANOVA indicated that this effect of Limb was not statistically significant (BBB: $F_{1,9} = 2.29$, $p = 0.17$; TBL: $F_{1,9} = 3.40$, $p = 0.10$).

Table 2.3: Mean and SE values of the intensity of mean neuromuscular activity (µV) for the BBB and the TBL muscle of the impaired and the less-impaired limb presented for each viewing condition (ViewImp, ViewLessImp).

BBB	ViewImp	ViewLessImp
Impaired limb		
Mirror	29.9 ± 4.2	27.4 ± 5.7
Screen	27.9 ± 4.2	27.3 ± 5.6
Glass	31.0 ± 6.3	30.6 ± 5.2
Less-impaired limb		
Mirror	18.2 ± 3.8	16.2 ± 3.2
Screen	17.6 ± 3.4	21.3 ± 4.4
Glass	17.5 ± 4.5	26.2 ± 7.2

TBL	ViewImp	ViewLessImp
Impaired limb		
Mirror	12.4 ± 2.2	13.9 ± 3.5
Screen	12.4 ± 2.0	17.3 ± 5.4
Glass	15.4 ± 4.3	16.8 ± 3.9
Less-impaired limb		
Mirror	7.3 ± 1.1	8.4 ± 1.4
Screen	8.8 ± 1.3	8.8 ± 1.4
Glass	6.8 ± 1.1	10.6 ± 1.9

Relative duration of concentric and eccentric activity in the BBB muscle

No significant main or interaction effects were found for the concentric activity of the BBB muscle (see Table 2.4). Mirror visual feedback, irrespective of which arm was viewed, did not have an effect on the relative contribution of concentric BBB activity to execution of the movement in the impaired or less-impaired arm ($F_{2,18} = 0.36$; $p = 0.70$). Additionally, there tended to be more concentric activation in the impaired limb than in the less-impaired limb (25.8 ± 3.9% vs. 17.2 ± 4.4%), but this difference was insignificant ($F_{1,9} = 2.74$, $p = 0.13$).

For the eccentric activity of the BBB muscle a significant main effect of Limb was found ($F_{1,9} = 7.53$, $p = 0.02$) with the impaired limb having 16.3% more eccentric activity than the less-impaired limb. This effect was accompanied by a three-way interaction between Limb, Viewing side and Visual condition ($F_{2,18} = 4.67$, $p = 0.02$). Figure 2.2 illustrates this interaction using the difference in eccentric activity between the two viewing sides (i.e. ViewImp and ViewLessImp) for the impaired and less-impaired limb and for each visual condition. This *difference score* was determined by subtracting the eccentric activity in the ViewImp condition from the eccentric activity in the ViewLessImp condition. A negative difference score then indicates lower eccentric activity in the ViewLessImp condition whereas a positive difference score represents higher eccentric activity in the ViewLessImp condition. Inspection of Figure 2.2 and post-hoc examination of the three-way interaction indicated that there were no effects of Visual condition or Viewing side on the eccentric activity of the less-impaired arm. For the impaired arm, however, mirror visual feedback from the impaired arm resulted in 10.3% more eccentric activity than mirror visual feedback from the less-impaired arm (p = 0.007). Furthermore, a significant effect of Viewing side was also present in the glass condition, where looking from the less-impaired side resulted in more eccentric activity than looking from the impaired side (mean difference score = 8.7%, p = 0.02). Viewing side did not have an effect on the eccentric activity of the BBB in the screen condition. Finally, focusing on the differences in eccentric activity between the visual conditions (see Table 2.4) it was found that mirror visual feedback of the less-impaired arm resulted in less eccentric activity in the impaired arm than the glass condition when viewing from the same side (mean difference = 12.8%, p = 0.001). In addition, for the ViewLessImp condition, the glass condition was performed with more eccentric activity in the impaired arm than the screen condition (mean difference = 8.2%, p = 0.02).

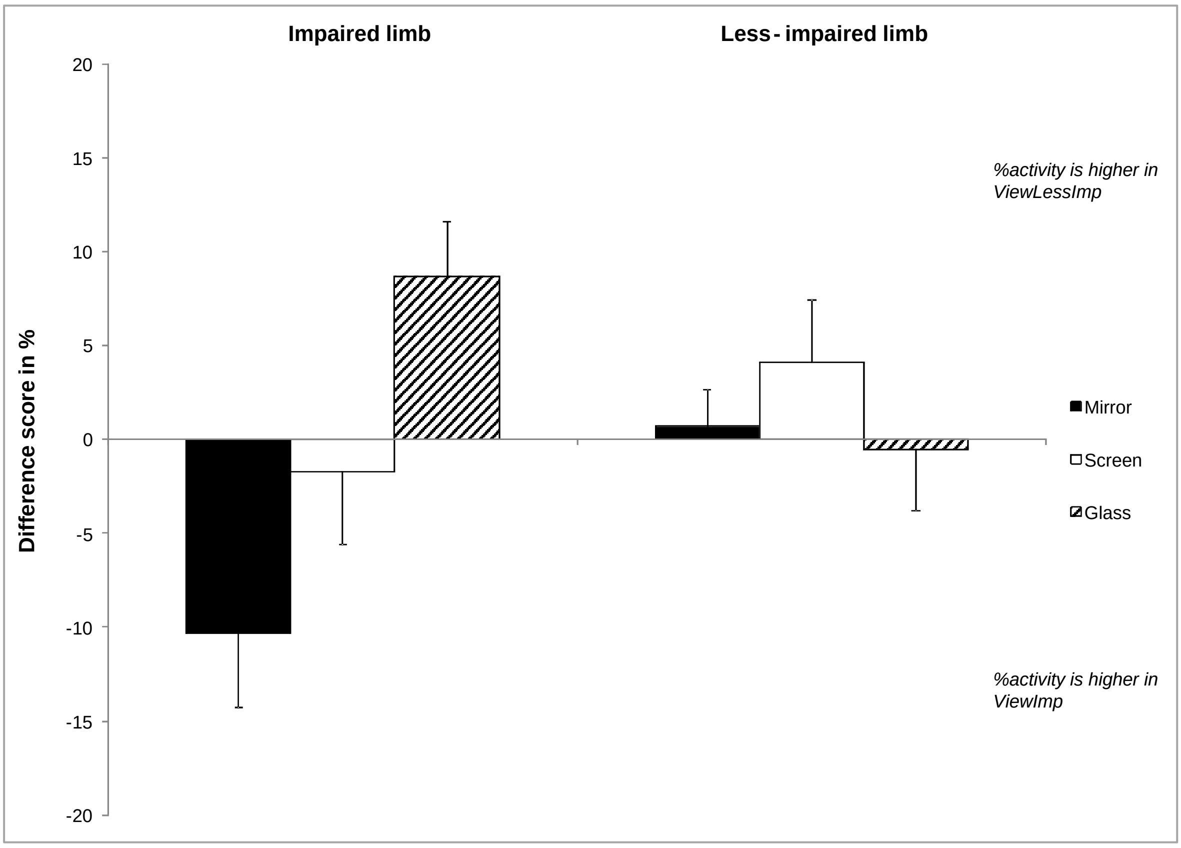

Figure 2.3: Difference scores of the relative duration of eccentric activity (in percentage) in the BBB muscle of the impaired (left side of the figure) and the less-impaired limb (right side of the figure) for the mirror (black bars), screen (white bars), and glass (dashed bars) condition. A positive difference score means that the eccentric activity is higher in the ViewLessImp compared to the ViewImp condition and a negative difference score means that the eccentric activity is lower in the ViewLessImp condition compared to the ViewImp condition.

Relative duration of concentric and eccentric activity in the TBL muscle

For the concentric activity of the TBL muscle a significant interaction effect between Limb and Viewing side was found ($F_{1,9}$ = 10.47, p = 0.01; see Table 2.4). The concentric activity in the impaired limb was larger than in the less-impaired limb for both the ViewImp and the ViewLessImp condition (mean difference = 8.56% and 4.56%, respectively). Furthermore, viewing from the less-impaired side resulted in longer durations of concentric activity in the less-impaired limb than viewing from the impaired side, irrespective of the visual condition (mean difference = 3.49%). For the eccentric activity of the TBL, no effect of Limb, Visual condition, or Viewing side was found.

Table 2.4: Mean and SE values of the eccentric and concentric muscle activity, expressed as a percentage of the total movement, of the Biceps Brachii Brevis (BBB) and the Triceps Brachii Longus (TBL) in the impaired and less-impaired limb for theViewImp (viewing the movement from the impaired side of the body) and ViewLessImp (viewing the movement from the less-impaired side of the body) conditions.

		BBB (%muscle activity)		
	Eccentric		**Concentric**	
	ViewImp	ViewLessImp	ViewImp	ViewLessImp
Impaired limb				
Mirror	34.2 ± 4.9	23.9 ± 6.5	26.6 ± 3.7	26.1 ± 4.2
Screen	30.2 ± 5.5	28.5 ± 7.2	25.7 ± 4.7	22.5 ± 3.6
Glass	28.0 ± 6.1	36.7 ± 6.3	25.1 ± 5.4	28.6 ± 4.1
Less-impaired limb				
Mirror	12.5 ± 4.1	13.2 ± 4.5	16.4 ± 5.1	16.2 ± 4.5
Screen	12.2 ± 4.1	16.3 ± 4.3	17.4 ± 5.0	18.8 ± 4.6
Glass	15.1 ± 5.6	14.5 ± 3.7	16.2 ± 5.3	18.3 ± 5.2

		TBL (%muscle activity)		
	Eccentric		**Concentric**	
	ViewImp	ViewLessImp	ViewImp	ViewLessImp
Impaired limb				
Mirror	7.3 ± 2.8	11.6 ± 4.2	10.5 ± 3.7	9.9 ± 4.9
Screen	9.1 ± 3.4	11.7 ± 4.0	11.8 ± 3.4	13.5 ± 5.2
Glass	10.8 ± 4.6	13.0 ± 4.8	12.7 ± 4.5	13.0 ± 4.7
Less-impaired limb				
Mirror	3.4 ± 1.6	4.9 ± 2.3	1.7 ± 0.7	3.8 ± 1.4
Screen	5.2 ± 1.8	3.2 ± 1.2	4.3 ± 1.5	5.7 ± 2.0
Glass	2.2 ± 1.5	8.3 ± 2.6	1.8 ± 1.2	8.8 ± 3.0

Discussion

This study investigated the effect of mirror visual feedback from the impaired arm (‘compromised’) compared to mirror visual feedback from the less-impaired arm (‘uncompromised’) on the interlimb coupling and the neuromuscular control during a bimanual coordination task in children with SHCP. In doing so, we wanted to determine whether previously found effects of the mirror box illusion in these children (Feltham, Ledebt, Bennett et al., 2010; Feltham, Ledebt, Deconinck et al., 2010b) were the result of the mirror and the related perception of visual symmetry per se or of the illusion that the impaired arm appears to move with less jerk and in synchrony with the less-impaired arm.

While the former would mean that ‘compromised’ as well as ‘uncompromised’ mirror visual feedback can trigger an improvement of the bimanual coupling and/or the neuromuscular activation, the latter can only be elicited by ‘uncompromised’ mirror visual feedback.

The CRP, which gives an indication of the nature of the bimanual coupling during this task, i.e. the synchronicity of the two limbs, indicates that the less-impaired arm was ‘leading’ the impaired arm across all conditions. This is in congruence with earlier studies on bimanual coordination in typically developing children (Pellegrini, Andrade, & Teixeira, 2004) and adults (e.g. Amazeen, Amazeen, Treffner, & Turvey, 1997; Stucchi & Viviani, 1993; Treffner & Turvey, 1995). The asynchrony of approximately 10° falls within the higher range of previously reported values in children with SHCP (Feltham, Ledebt, Bennett et al., 2010: -0.3°; Volman et al., 2002.: -5° to 9°), but is still acceptable given the unilateral impairment of the children. Note that the phase lag between the two hands may indicate that the movement of the lagging impaired hand may be guided by visual feedback from the less-impaired hand. However, the CRP did not change as a function of visual condition or viewing side, which suggests that the bimanual coupling is clearly not solely governed by a visual feedback mechanism and that processes relying on central representations of action do contribute to the coupling as well (addressed below).

It thus seems that mirror visual feedback did not influence the interlimb coupling and there was no difference between ‘compromised’ and ‘uncompromised’ mirror visual feedback. Interestingly, however, the mirror did have an effect on the neuromuscular activity required to perform the task. This suggests that, although the movement performance itself remained the same, the muscular effort responsible for this movement did change in response to the available visual information. Our results demonstrate that mirror visual feedback led to a reduction of eccentric BBB activity in the impaired arm compared to the glass condition and, importantly, this effect was exclusive to ‘uncompromised’ mirror visual feedback, i.e. viewing the less-impaired arm and its mirror reflection (ViewLessImp). In the impaired arm, mirror visual feedback of the less-impaired arm appears to have the capacity to improve the neuromuscular efficiency of the impaired arm by reducing the disproportionally high eccentric activity. The finding that ‘compromised’ mirror visual feedback did not elicit a similar effect, shows that the mirror effect in children with SHCP is not just a response to the visual symmetry, but is also dependent on the type of visual information generated by the mirror. The latter nuances the findings of Franz and Packman (2004) who found that mirror visual feedback enhanced the bimanual coupling (i.e. similarity in range of motion of the two hands) in typical adults, irrespective of viewing mirror feedback from the left or the right hand. However, unlike in typical adults, in children with SHCP the nature of mirror visual feedback from the left and right hand is qualitatively different, which might explain the apparent discrepancy between the two studies.

The finding from the present study that mirror visual feedback of the impaired arm has the opposite effect of 'uncompromised' apparent symmetrical motion in children with SCHP, qualifies the findings of Feltham, Ledebt, Deconinck et al. (2010b) who only looked at the effect of mirror feedback from the less-impaired arm. We demonstrated that the favourable results (i.e. the reduction in eccentric BBB activity in the impaired arm) are not just due to the visual perception of apparent bimanual symmetry per se. Instead children with SHCP appear to benefit specifically of mirror visual feedback from the less-impaired arm, which seems to be in line with the notion of Ramachandran (2005). Ramachandran hypothesised that mirror visual feedback may assist the central control of movement in people with unilateral motor problems by restoring the congruence between disrupted sensory information and the central motor command signals. According to this view, the information provided by the mirror could assist in the neuromuscular control of the movement by replacing conflicting visual feedback of the impaired limb with feedback that is in accordance with the intended movement (i.e. 'uncompromised' visual feedback of the less-impaired limb). By showing that the mirror-effect on motor performance in children with SHCP is specifically related to mirror visual feedback of the less-impaired arm, the current study provides a valuable contribution to the discussion about the underlying mechanisms of this effect. Nevertheless, the actual neural underpinnings will only be revealed using advanced neuro-imaging techniques. In addition, it may be surprising that a short exposure to the mirror already induces these effects on the neuromuscular activity and future studies should examine the impact of longer exercise or interventions with mirror feedback. Related to this issue is the fact that no (major) effect of the mirror was observed on the bimanual coupling or neuromuscular measures such as the intensity of mean neuromuscular activity, the eccentric activity in the TBL muscle, and concentric activity in the BBB muscle. Furthermore, we cannot exclude the limited number of trials (three per condition) and the large age range of the participants to affect the precision and generalization of the results. The precision of the measurement might be enhanced with larger number of trials, but in the current study it was high enough to reveal significant differences between the conditions. One can expect that a larger number of trials will enhance the actual results but one must also consider that the limited attention span and fatigability of the participants with cerebral palsy might interfere. Considering that the present study used a repeated measures design each participant was his own control and the variability that the large age range may have introduced was nevertheless small enough to show a significant effect of the experimental conditions. While we did not anticipate an age effect, we cannot exclude it and suggest that this should be further investigated.

In conclusion, this study provided more insight into the effects of mirror visual feedback in children with SHCP. We showed that the effects found by Feltham, Ledebt, Bennett et al. (2010) and Feltham, Ledebt, Deconinck et al. (2010b) on neuromuscular activity and bimanual coordination, are likely not caused by the perception of two symmetrically moving limbs per se. Instead, for an increase in neuromuscular efficiency of bimanual movement (i.e. a decrease in excessive eccentric activity in the arm flexors), children with SHCP require mirror visual feedback of the ('unaffected') less-impaired limb.

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Chapter 3

Deficits in upper limb position sense of children with Spastic Hemiparetic Cerebral Palsy are distance dependent

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Abstract

This study examined the arm position sense in children with Spastic Hemiparetic Cerebral Palsy (SHCP) and typically developing children (TD) by means of a contralateral matching task. This task required participants to match the position of one arm with the position of the other arm for different target distances and from different starting positions. Results showed that children with SHCP exhibited with both arms larger matching errors than the TD group, but only when the distance between the arms at the start of the movement was large. In addition, the difference in errors between the less-impaired and the impaired limb changed as a function of the distance in the SHCP group whereas no interlimb differences were found in the TD group. Finally, spasticity and restricted range of motion in children with SHCP were not related to the proportion of undershoot and size of absolute error. This suggests that SHCP could be associated with sensory problems in conjunction with their motor problems. In conclusion, the current study showed that accurate matching of the arms is greatly impaired in SHCP when compared to TD children, irrespective of which arm is used. Moreover, this deficit is particularly present for large movement amplitudes.

Introduction

Proprioception refers to the sense of body parts in space and comprises a static (sense of static limb position or position sense) and a dynamic component (sense of movement or kinaesthesia). It is a complex somatosensory modality that is imperative for the control of movement.

A large body of evidence details the critical role of proprioception in controlling muscle interaction torques (e.g. Sainburg, Ghilardi, Poizner, & Ghez, 1995) in timing the coordination between limb segments (Cordo, Carlton, Bevan, Carlton, & Kerr, 1994), in monitoring movement trajectories (Ghez, Gordon, Ghilardi, Christakos, & Cooper, 1990), and in establishing internal representations used during the acquisition and adaptation of skilled movement (Kawato & Wolpert, 1998). It is therefore not surprising that impaired proprioception is often suggested to be implicated in motor dysfunction such as in Parkinson’s disease (Adamovich, Berkinblit, Hening, Sage, & Poizner, 2001), hemiparetic stroke (Niessen et al., 2008), cerebellar disorders (Cody, Lovgreen, & Schady, 1993) or cerebral palsy (CP; Cooper, Majnemer, Rosenblatt, & Birnbaum, 1995; Opila-Lehman, Short, & Trombly, 1985). Still, to facilitate the design of tailored therapeutic interventions, empirical research is required to get a detailed and more complete view of the deficits encountered by disabled individuals.

A number of studies have already shed light on proprioception in CP. CP is a group of permanent disorders of movement and posture due to a non-progressive lesion in the foetal or infant brain (Miller, 2007). In children with Spastic Hemiparetic CP (SHCP) impaired control of muscle tone and spasticity in the limbs on one side of the body (the impaired side) severely complicates normal daily movement function. These deficits in daily functioning become predominantly evident for movements executed with the arm, which is usually more affected than the lower extremity (Charles & Gordon, 2006). Goble, Hurvitz, and Brown (2009) examined joint-position sense in this population using an arm flexion/extension task. This task required the participants to match the position of the elbow (occluded from view) to a target position to which the elbow had been extended passively before the start of the trial. Larger errors were made with the impaired limb than with the less-impaired limb, and the latter was as accurate as the limbs of typically developing (TD) control children. It should be noted however, that in a sub-sample of the CP-population the condition is accompanied with memory deficits (Bottcher, 2010; Kolk & Talvik, 2000), which may have contributed to the reduced ability to match a previously felt position and complicates the interpretation of the results. Indeed, the contrasting findings of Chrysagis, Skordilis, Koutsouki, and Evans (2007) who showed with a similar task that children with SHCP made significantly larger errors than TD children with the impaired as well as the less-impaired arm, might be due to differences in the children's ability to memorize positions. Wingert, Burton, Sinclair, Brunstrom and Damiano (2009) used an alternative approach and tested joint-position sense using a forearm pronation/supination task in which the position of the occluded hand was to be aligned with a visual target. The 'cross-modal matching' required in this task, i.e. mapping between visual and proprioceptive information, adds another degree of difficulty (e.g. von Hofsten & Rosblad, 1988; Wann, 1991) and again implies that this task cannot be completed using somatosensory information only. In agreement with other work, this study showed that larger errors were made with the impaired limb than with the less-impaired limb. However, the overall performance of the hemiplegic group did not differ from the control group. Taken together, it thus seems that the accuracy of the joint-position sense (and the associated proprioceptive cues) is dependent on the joint (and the related muscle group) tested. In addition, these studies illustrate that it is difficult to assess joint-position sense in isolation (i.e. without confounding factors such as memory load or multi-modal mapping). Still, one aspect of joint-position sense that has not been considered in the study of SHCP is the ability to match the position of limbs in a contralateral matching task where the participant is instructed to copy the position of one limb by placing the other, contralateral limb, in the same mirror symmetric position. Such an intra-modal matching test, which does not require re-mapping between sensory inputs and in which the involvement of memory is considerably reduced, can provide us with useful information about how problems with proprioception influence tasks that involve both arms. This is particularly relevant for the study of children with SHCP whose motor impairments appear to be limited to one body side, but are known to hamper bimanual actions (Charles & Gordon, 2006). Therefore, in this study we will explore to what extent matching movements, in which both hands are involved, are hindered in children with SHCP by means of a contralateral matching task.

It has been suggested that position sense is dependent on the location (relative to the body) at which the measurement is performed. Localization of the hand is more precise in proximity of the body (i.e. at smaller distances relative to the body) than at larger distances from the body (van Beers, Sittig, & Denier van der Gon, 1998; Wilson, Wong, & Gribble, 2010). This phenomenon has been reported in studies of young (Goble & Brown, 2008; Goble, Lewis, & Brown, 2006) and elderly (Adamo, Martin, & Brown, 2007), supporting the notion that this effect is common and probably robust against neurodegeneration. Van Beers et al. (1998) suggested that better localization at distances closer to the body may be understood from the geometry of the arm, alongside anatomical and physiological properties such as the fact that the number of muscle spindles acting about the joints in the arm increase in proximal direction (Scott & Loeb, 1994 In: van Beers et al., 1998). Verifying whether the accuracy in a proprioceptive-guided matching task in children with SHCP follows a similar trend (i.e. decrease in precision for locations further away from the body) may thus serve to test whether they are subject to similar anatomical and physiological constraints and use similar cues to localize the position of their hands as compared to TD children. To the best of our knowledge, this aspect has been largely overlooked in previous research into position sense of children with SHCP.

The aim of this study was therefore to add to the existing body of knowledge on proprioception in children with SHCP, and more specifically to gain insight into the accuracy of position sense of the impaired and less-impaired arm in a contralateral matching task. In a case study (N=2) using a similar task Lee, Daniel, Turnbull, and Cook (1990) found that children with SHCP experienced difficulties with matching for both the impaired and less-impaired arm. The purpose of the current study was to substantiate these findings. In addition, considering the location-dependent effect on position sense, this study aimed to examine whether the accuracy of matching performance and possible differences between the SHCP and TD group on a contralateral matching task are location-dependent (i.e. dependent on the distance relative to the body). If the distance effect in children with SHCP does not significantly deviate from TD children, this could suggest that both groups use similar sensory cues to localize the hand and are subject to similar anatomical and physiological constraints, despite possible disturbances in the input and/or processing of sensory information.

Methods

Participants

Fourteen children with SHCP participated in this study (mean age 12.5 ± 1.9 years) of which six had a right and eight had a left hemiplegia (see Table 3.1 for further details). The participants were free from any neuromuscular disorders other than CP, did not have visual impairments or pain in either of the upper limbs, and they were not treated with Botulinum toxin in the past six months preceding the measurement. The children with SHCP were recruited through the Dutch society for children with a physical handicap and their parents. Before the actual start of the experiment, the Manual Ability Classification System (MACS), Functional Independence Measure (WeeFIM) and Tardieu score for spasticity were defined for the SHCP group in order to get an indication of the severity of the disorder (Table 3.1). The MACS describes how children use their hands during object handling and their need for assistance to perform manual skills in everyday life (Eliasson et al., 2006). The severity of performance limitation and the degree of required assistance increases for each MACS level from 1 to 5. Seven children were classified in MACS level 3, five children in level 2 and two children in level 1. The WeeFIM scores range from 13 to 91 with a higher score representing a better functional independence. In the current population the WeeFIM scores ranged from 52 to 91. Finally, the Tardieu score was determined by a qualified physiotherapist as an indication of the children's spasticity level. Individual scores were measured for the Biceps Brachii Brevis and the Triceps Brachii Longus and combined into one total score. All children showed mild to moderate spasticity with Tardieu scores ranging from 0.5 to 2.

In addition, a reference group of twenty TD children without any history of neuromuscular disorders and within the same age range as the children with SHCP (mean age 12.9 ± 2.6 years) were recruited among the university staff's families and friends. The TD children all had normal or corrected-to-normal vision and all but one were right hand dominant (determined by means of the Edinburgh Handedness Inventory; Oldfield, 1971). Participant characteristics can be found in Table 3.1 (SHCP) and Table 3.2 (TD). Prior to testing the participant's parents provided written informed consent. All procedures were approved by the institutional research ethics committee and were in accordance with the Declaration of Helsinki.

Materials and procedure

The child was seated on a height adjustable chair without armrests at a height adjustable table with the knees 90° flexed. Position sense was assessed using a custom made device consisting of two handles, each on a separate track fixed to a horizontal panel. The tracks were 20 cm apart, parallel to each other, and perpendicular to the medio-lateral axis of the trunk. The children were positioned such that the centre of the body was located in between the two tracks, and with the beginning of the track at 15 cm from the upper body. Vision of the limbs was blocked with an opaque cover on top of the wooden construction. The experimental setup is depicted in Figure 3.1. The position of two parallel handles outside the box was recorded using one Optotrak unit with three infrared cameras (3020 Optotrak, Northern Digital Inc., Waterloo, Canada), which enabled us to calculate the position of the hands inside the box.

Before the start of the actual experiment, the maximum reaching distance of both arms was determined (MRD) in order to scale the different matching positions across subjects. MRD corresponds to the distance from the start of the track (position most proximal to the body) to the position of the handles when the elbows were extended as far as possible without bending the trunk forward. The MRD was used to determine the three target positions to be tested in the matching task, i.e. 25%, 50%, and 75% of the MRD. In case the MRDs of the left and right arm were different, the three target positions were based on the smallest MRD (this was applied for both groups). This means that for the children with SHCP the target positions were always based on the MRD of the impaired arm. The MRDs for each individual are reported in Table 3.1 and 3.2.

Table 3.1: Participant characteristics of the SHCP group. For each participant the age in years, sex, dominant hand, WeeFIM score, MACS level, Tardieu Scale, aetiology, and the Maximum Reaching Distance (MRD) for the dominant and non-dominant arm are presented.

P	**Age (years)**	**Sex**	**Dominant arm**[a]	**WeeFIM/MACS**	**TS**[b]	**Aetiology**	**MRD D/ND**[c]
1	13.4	M	Right	78/3	2	O_2 shortage during birth	41/27.2
2	10.5	M	Right	88/3	2	Cerebral infarction	47/30
3	10.8	M	Right	91/2	1.5	Unknown	33/31.5
4	14.5	M	Right	62/3	2	Schizencephaly	48/36.5
5	13.6	M	Right	91/2	2	Cerebral infarction	34/31.5
6	10.8	F	Right	52/3	1.5	Cerebral haemorrhage	31/26
7	12.1	F	Left	91/3	1	Cerebral infarction (thalamus)	46/42
8	15.5	M	Left	76/1	2	Unknown	47/46.5
9	9.3	M	Left	91/1	1	Cerebral infarction	25.5/24.5
10	13.1	F	Left	91/2	2	Cerebral infarction	39/38
11	14.4	M	Left	81/2	1	Cerebral haemorrhage	33.5/24.5
12	12.5	M	Left	59/3	2	Cerebral infarction	34/22.2
13	14.3	M	Left	71/3	2	Unknown	38/36.5
14	10.6	M	Left	87/2	0.5	O_2 shortage during birth	31/30.3

[a] *The dominant arm is the less-impaired arm.*

[b] *Tardieu Score = mean of the individual scores of the Biceps and the Triceps.*

[c] *MRD = Maximum Reaching Distance; D = dominant/less-impaired limb; ND = non-dominant/impaired limb.*

Table 3.2: Participant characteristics of the TD group. For each subject the age in years, sex, dominant hand, score of the Edinburgh Handedness Inventory, and the Maximum Reaching Distance (MRD) for the dominant and non-dominant arm are depicted.

P	**Age (years)**	**Sex**	**Dominant hand**	**EHI score**[a]	**MRD D/ND**[b]
1	13.0	M	Right	100	42/41
2	13.2	F	Right	100	37/37
3	12.3	F	Right	100	33/35
4	13.4	M	Right	100	36/34.5
5	8.3	F	Right	89	30/29
6	10.0	F	Right	80	30.5/29.5
7	16.9	F	Right	100	33.5/32.5
8	12.9	F	Right	90	34/33
9	13.3	F	Right	90	36/34
10	15.1	M	Right	90	40/40
11	11.4	M	Right	50	36/37
12	16.3	F	Right	40	32.5/34
13	10.9	F	Right	70	32.5/32.5
14	12.1	F	Right	60	38/37
15	16.5	F	Right	100	42/42
16	17.4	F	Right	70	35.5/34.5
17	14.9	M	Right	70	34/34
18	10.6	F	Right	100	28/27
19	10.6	M	Right	100	40/40
20	10.1	F	Left	-50	31/30

[a] *EHI score = Edinburgh Handedness Score. +100 is complete right handedness; -100 is complete left handedness. If EHI was between -50 and +50 (ambidexter), the writing hand was identified as the dominant hand.*

[b] *MRD = Maximum Reaching Distance; D = dominant limb; ND = non-dominant limb.*

The contralateral matching task required participants to match the position of one limb (reference limb), which was moved to the predetermined target position passively, by actively moving the other limb (matching limb) to the (mirror symmetric) position at the same distance as the reference arm. Three target positions (25%, 50%, and 75% of the MRD) were tested and the matching was done with either the less-impaired limb (dominant for TD children) or the impaired limb (non-dominant for TD children). The matching limb started at MRD (distally) or at the beginning of the track (proximally). The combination of all independent variables (3 target positions of the reference limb, 2 matching limbs, and 2 start positions of the matching limb), resulted in 12 trial types. Each trial type was performed once. The total amount of trials was divided in two blocks: 1) matching with the impaired (non-dominant) arm, 2) matching with the less-impaired (dominant) arm. The order of blocks was randomized over participants and within each block the order of the trial types was randomized to reduce possible thixotropic effects on the matching accuracy (Proske, 2006). Prior to data collection 3 practice trials were conducted to familiarize the participant with the test setup and to check if the children were able to perform the movement properly. If the participant was unable to grip the handle due to his/her physical impairment, the experimenter placed the hand on top of the handle. However, in none of the participants the handle slipped out of the hands during a trial. In order to keep the children motivated they were told that the better their performance the more points they would earn. At the end of the experiment they could trade their points for a small gift.

Data analysis

The position data of the reference and the matching limb were imported into Matlab (version 7.1, The Mathworks Inc.). Then, absolute endpoint error was determined as the distance between the two handles at the end of the movement using custom-written routines. The end of the movement was verified by visual inspection of the plot showing the time series of the matching limb's position (inter-rater reliability $r = 0.98$, $p < 0.001$).

In addition, we calculated the proportion of trials in which the matching arm overshot or undershot the position of the reference target, resulting in amplitudes that were larger or smaller than the actual reaching distance respectively.

Statistical analysis

The MRDs of the SCHP group and the TD group were compared with a two-way repeated measures ANOVA with Limb (dominant/less-impaired, non-dominant/impaired) as a within factor and Group (SHCP, TD) as a between factor. The endpoint error in the contralateral matching task was analysed using a four-way repeated measures ANOVA with Limb (non-dominant/impaired, dominant/less-impaired), Position of the reference limb (25%, 50%, 75% MRD; i.e. the distance relative to the body), and Start position (distal, proximal) as within subjects factors and Group (SHCP, TD) as a between subjects factor. In case the sphericity assumption was violated, Greenhouse-Geisser adjustments were made. Fishers' LSD was used for post hoc analysis. To compare the proportions of undershoots and overshoots, a non-parametric Mann-Whitney U test was performed on the relative number of undershoots between the TD and the SHCP group. The significance level was set at 0.05.

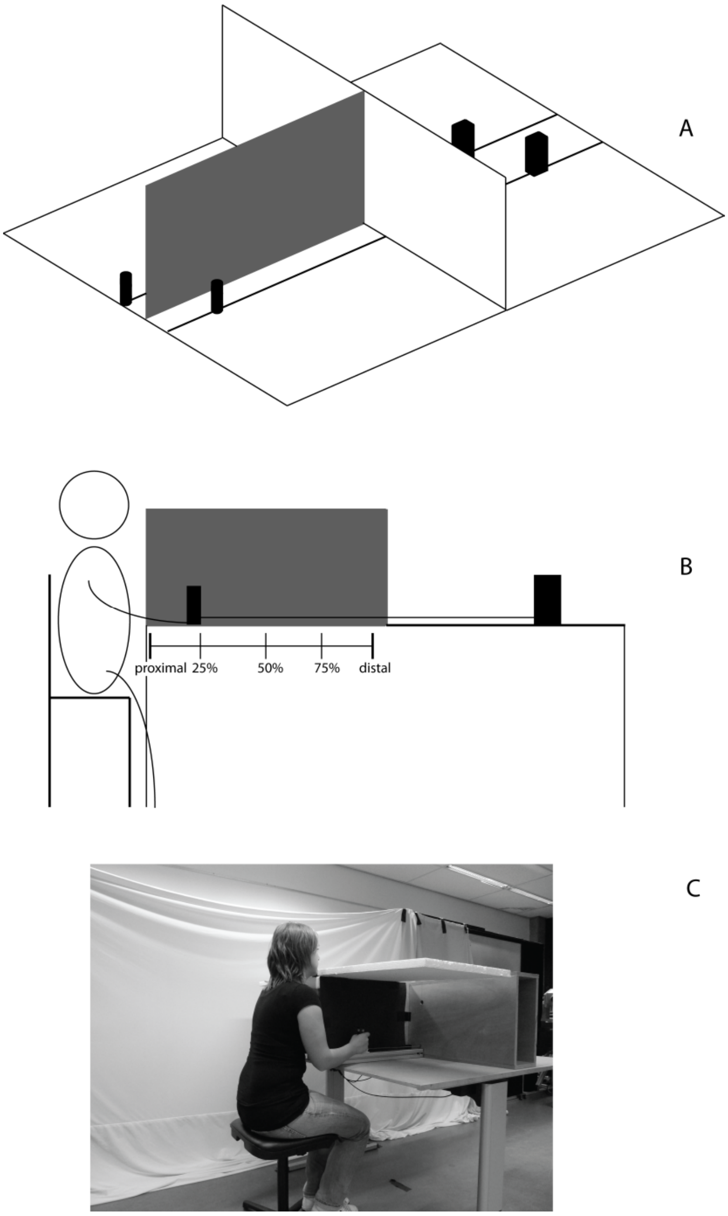

Figure 3.1: (A) Top view of the experimental setup with the two handles that could be slid back and forth along the track. The screen between the arms prevented the hands from touching each other. The position of the handles outside the box was measured with an Optotrak camera (not depicted here). In this picture the opaque cover on top of the construction is not visible.

(B) Side view of the experimental setup. The starting positions (proximal, distal) and the three target positions (25%MRD, 50%MRD, 75%MRD) are indicated. Please note that the target positions and the distal start positions (MRD) were determined based on the Maximum Reaching Distance of the child and thus differed per participant.

(C) Real-life picture of the experimental setup with an opaque cover on top of the construction.

Results

Maximum Reaching Distance (MRD)

A Limb x Group interaction ($F_{1,32} = 17.31$, $p < 0.001$) revealed that in children with SHCP the MRD of the less-impaired limb was larger than the MRD of the impaired limb

($p < 0.001$; 37.7cm vs. 31.9cm), while no such difference was found in TD children ($p = 0.63$; 35.1cm vs. 34.7cm, for dominant and non-dominant arm respectively). Further post-hoc analysis of the Limb x Group interaction did not show differences in MRD between the limbs of the SHCP group and the limbs of the TD group (Dominant arm: 37.7cm [SHCP] vs. 35.1cm [TD]; Non-dominant arm: 31.9cm [SHCP] vs. 34.7cm [TD]).

Endpoint error

All children were able to complete the experiment, but due to technical problems with the motion capture system during a number of trials of participants 7 (1 trial), 11 (2 trials), and 12 (2 trials) of the SHCP group, the data of these participants could not be included in the statistical analysis.

Analysis of the absolute error in the matching task revealed a two-way interaction between the factors Position reference and Start position ($F_{2,58} = 32.73$, $p < 0.001$), which was also present in two three-way interactions: Position reference x Start position x Group ($F_{2,58} = 5.26$, $p = 0.008$) and Position reference x Start position x Matching limb ($F_{2,58} = 3.29$, $p = 0.04$). Inspection of this Position reference x Start position interaction (see Figure 3.2) showed an almost symmetrical picture for trials starting at a distal point and trials starting in proximity of the body, for both groups. Absolute error at 25%MRD in trials starting in the proximity of the body (i.e. 0%MRD) was similar to the absolute error at 75%MRD in trials starting at the most distal point from the body (100% MRD). Likewise, absolute error at 75%MRD in trials starting proximal to the body (i.e. 0%MRD) was not different from absolute error at 25%MRD in trials starting at the most distal point from the body (100% MRD). Finally, a distal or proximal start of the matching limb did not affect the amplitude of the error when the reference limb was positioned at 50%MRD. In fact, this Position reference x Start position interaction reveals a Distance effect indicating gradually larger absolute errors for larger reaching distances, i.e. the distance that has to be covered by the matching hand in order to achieve an error of 0. A secondary 3-way repeated measures ANOVA (Limb x Distance x Group), in which the dependent variables Position reference and Start position were combined into one factor (Distance), yielded identical results as the initial 4-way ANOVA (Figure 3.3 explains the relation between the factors Position reference and Start position and Distance.) For reasons of clarity and comprehensibility, the results of the secondary analysis, in which all participants were included, will be presented here.

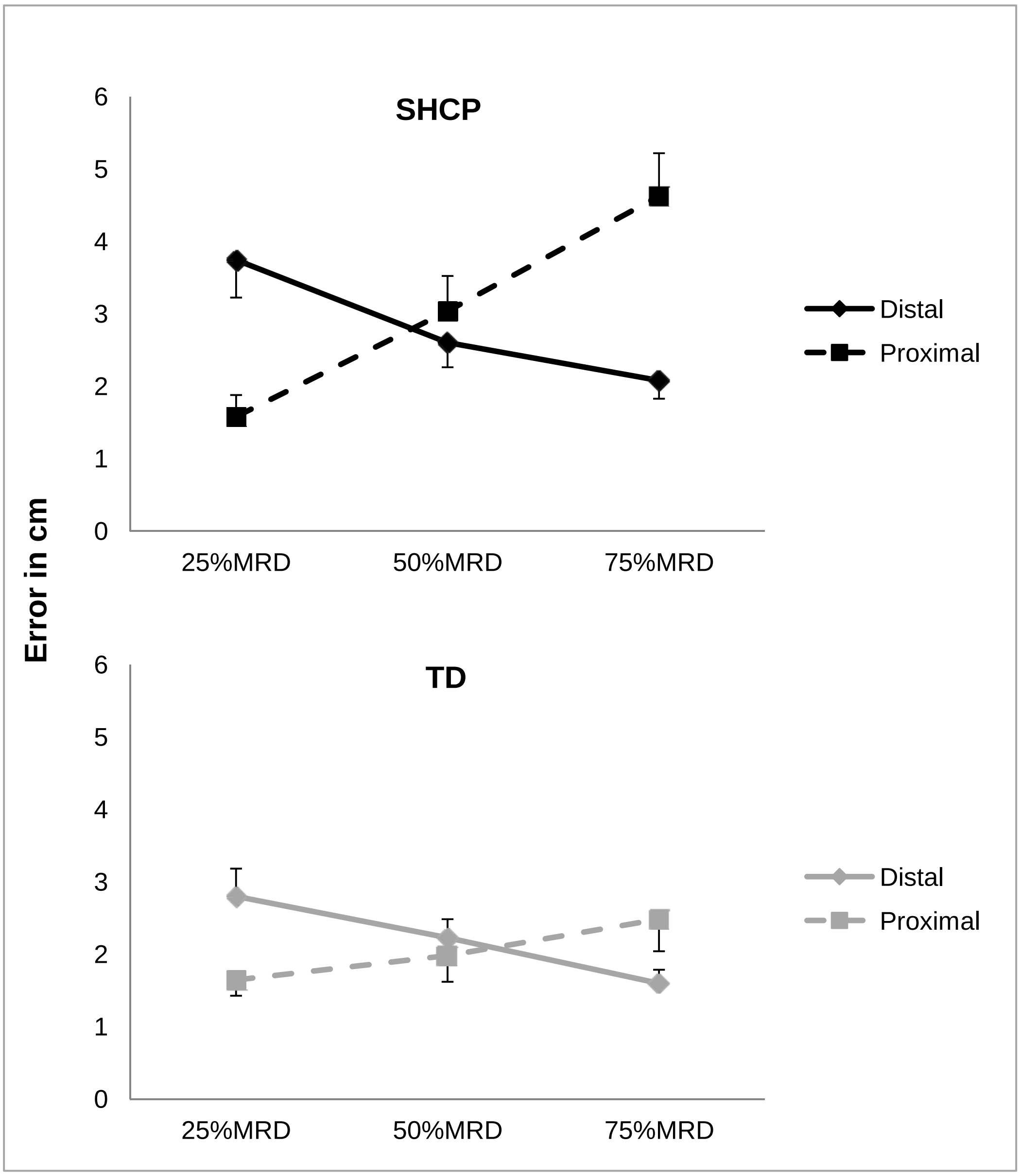

Figure 3.2: The absolute endpoint errors (in cm) on different positions of the reference limb (25%MRD, 50%MRD, 75%MRD) for the different starting positions (distal, proximal) for the SHCP (top graph) and the TD group. The solid line represents the errors when the matching position was at 25%, 50% or 75%MRD when starting the movement proximally to the body. The dashed line represent the errors when matching the arms at a target position at 25%, 50% and 75%MRD when starting the movement distally from the body.

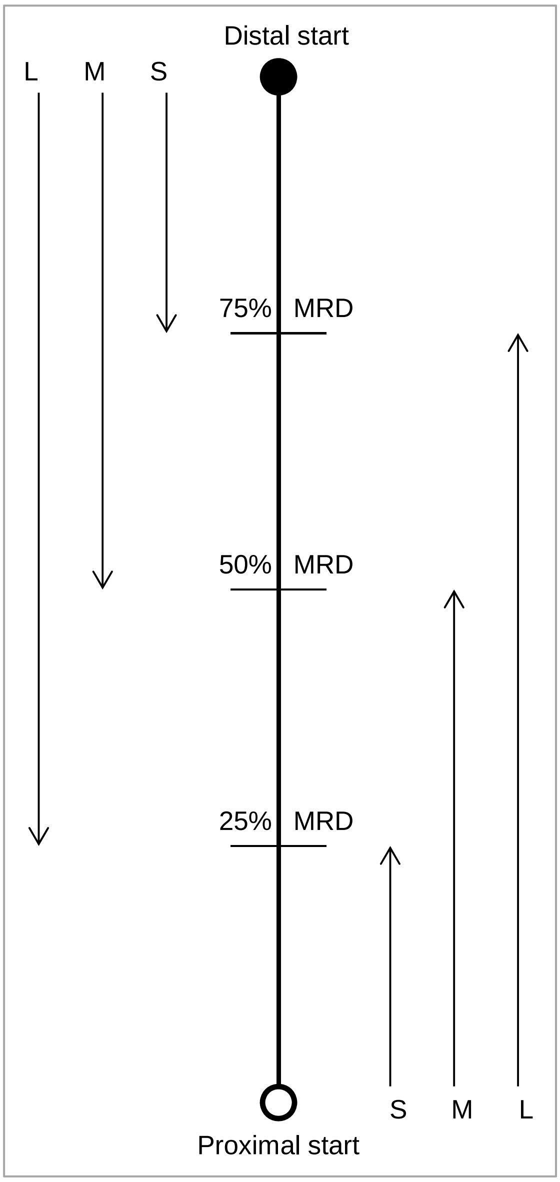

Figure 3.3: Conversion from Position reference (25%MRD, 50%MRD, 75%MRD) and Start position (proximal, distal) into Distance (small [S], medium [M], and large [L]). It can be seen that e.g. moving towards 25%MRD when starting proximally results in the same distance as moving towards 75%MRD when starting distally.

This secondary analysis revealed main effects of Group ($F_{1,32} = 72.41$, $p = 0.002$) and Distance ($F_{2,64} = 29.51$, $p = 0.002$) on absolute error, which were superseded by a Group x Distance interaction ($F_{1.4, 44.3} = 5.47$, $p = 0.006$; see Figure 3.4) and a Group x Distance x Limb interaction ($F_{2,64} = 3.78$, $p = 0.028$; see Figure 3.5). Post-hoc examination showed that the accuracy in this matching task dropped as a function of the reaching distance in both groups, but this drop in accuracy (i.e. increase in error) was significantly greater in the children with SHCP than in the TD children. This finding was further supported by the fact that there was no difference in absolute error between the SHCP and TD children for the small distance. In the medium distance the less-impaired limb of the SHCP group showed larger errors than the dominant arm of the TD group whereas no differences between the impaired arm and the non-dominant arm were found. Finally, when the reaching distance was large the errors made by both the impaired and the less-impaired arm were larger than in their counterparts of the TD group. Furthermore, no difference between the arms was found in TD children. In children with SHCP, however, matching with the impaired arm resulted in significantly larger absolute errors than matching with the less-impaired arm for the large distance condition (5.25cm vs. 3.99cm), while the opposite was found for the medium distance condition (2.64cm vs. 3.93cm). There was no difference between the impaired and less-impaired matching limb when the reaching distance was small.

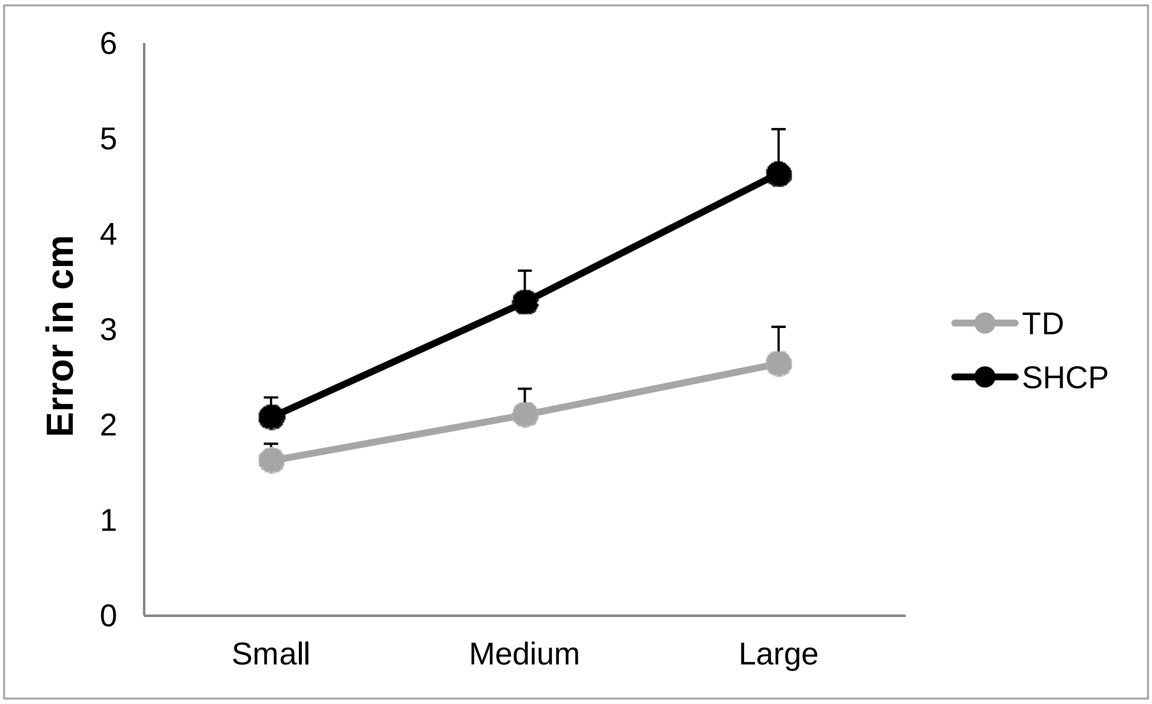

Figure 3.4: The absolute errors (in cm) of the typically developing (TD) and the Cerebral Palsy (SHCP) group for the different distances (small, medium, large).The black line represents the errors of the SHCP group and the grey line represents the errors of the TD group.

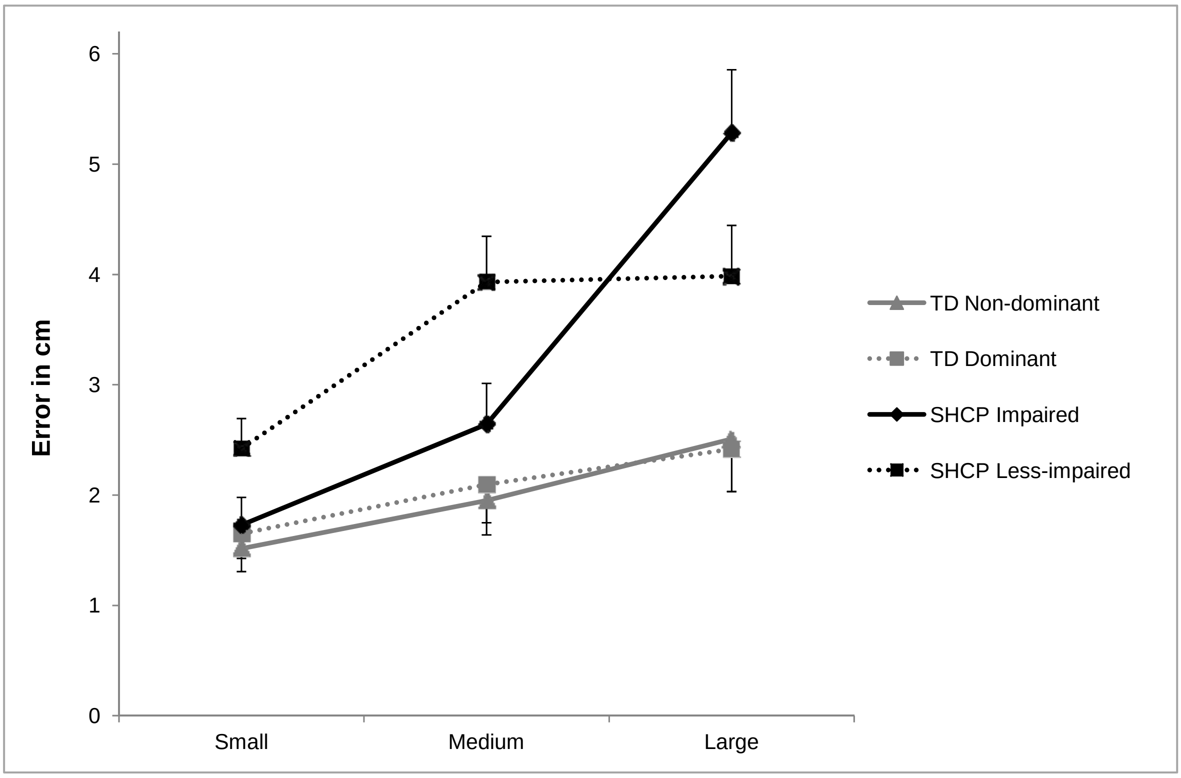

Figure 3.5: The absolute errors (in cm) of both upper limbs for the Typically Developing (TD) group (grey lines) and the Spastic Hemiparetic Cerebral Palsy (SHCP) group (black lines) depticted for each distance seperately. The distances (small, medium, large) are depicted on the horizontal axis. The solid grey line represents the errors of the non-dominant arm, the dashed grey line represents the errors of the dominant arm. The error of the impaired arm of the SHCP group are depicted with a solid black line and the error of the less-impaired arm is represented by the dashed black line.

Relative number of undershoot and overshoot

The proportion of trials resulting in an overshoot or undershoot is depicted in Table 3.3. All children undershot the target in the majority of the trials (TD: 80.8%, SHCP: 74.1%). These proportions were not significantly different (U = 103.0, z = -1.31, p = 0.19, average ranks = 19.4 and 14.9 for TD and SHCP respectively). In addition, inspection of Table 3.3 shows that the relative number of undershoots increased with increasing distance in both groups. The differences in the proportion of undershoots between the arms were small, especially in the SHCP group.

Table 3.3: Percentages (and proportions) of the overshoots and undershoots in the SHCP (impaired and less-impaired arm) and the TD group (non-dominant and dominant arm) in the small, medium and large distance. In the last column the total relative number of under- and overshoots is depicted. The range (in cm) of the total percentage overshoots (positive values) and undershoots (negative values) is indicated between brackets.

Undershoot		Small	Medium	Large	Total
SHCP	*Impaired*	66.7% (18/27*)	63% (17/27)	89.3% (25/28)	74.1% (-18.3 - -0.1)
	Less-impaired	73.1% (19/26)	71.5% (20/28)	81.5% (22/27)	
TD	*Non-dominant*	67.5% (27/40)	70% (28/40)	87.5% (35/40)	80.8% (-7.0 - -0.01)
	Dominant	80% (32/40)	87.5% (35/40)	90% (36/40)	
Overshoot					
SHCP	*Impaired*	33.3% (9/27**)	37% (10/27)	10.7% (3/28)	25.9% (0.03-5.7)
	Less-impaired	26.9% (7/26)	28.5% (8/28)	18.5% (5/27)	
TD	*Non-dominant*	32.5% (13/40)	30% (12/40)	12.5% (5/40)	19.2% (0.02-3.3)
	Dominant	20% (8/40)	12.5% (5/40)	10% (4/40)	

* Number of trials with undershoot/total number of trials

** Number of trials with overshoot/total number of trials

Relation with the level of spasticity and MRD

Two additional analyses were performed in order to examine whether the level of spasticity (Tardieu score) and the difference in MRD between the limbs have an influence on the magnitude of the absolute errors and on the number of trials with undershoot in children with SHCP.

For the first additional analysis, the children with SHCP were divided into two groups based on their spasticity level as indicated by the Tardieu score. One group ('mild spasticity group') included all children with a Tardieu score equal to or below 1 (n = 4) and the other group ('moderate spasticity group') included the children with a score above 1 (n = 10). The results of the Mann-Whitney U test revealed that the 'mild spasticity group' did not differ significantly from the 'moderate spasticity group' on the percentage undershoots (U = 15.5, z = -0.65, p = 0.51, average ranks = 8.6 and 7.0 respectively). Likewise, no differences between the group with scores equal to or below 1 and the group with scores above 1 were found for the absolute error when matching with the impaired limb on all three distances (Small: U = 14.0, z = -0.85, p = 0.39, average ranks = 6.0 vs. 8.1; Medium: U = 13.0, z = -0.99, p = 0.32, average ranks = 9.3 vs. 6.8; Large: U = 10.0, z = -1.14, p = 0.16, average ranks = 10.0 vs. 6.5).

For the second additional analysis, we compared the children with SHCP based on the relative difference of MRD between the less-impaired and the impaired arm. For each individual, the difference between the two MRDs (see Table 3.1) was divided by the largest MRD (expressed as a percentage) in order to minimize the inter-individual variability in arm length. The first group included the children with less than 10% relative difference (n = 8) and the second group included children with more than 10% relative difference (n = 6).

When comparing these two groups on relative number of undershoots, the Mann-Whitney U test did not reveal a significant difference between the groups (U = 14.5, z = -1.26, p = 0.21). The ‘more than 10% group’ showed an average rank of 5.9 and the ‘less than 10% group’ had an average rank of 8.7. This then suggests that both groups did not significantly differ on relative numbers of undershoot.

Also when focusing on the absolute error, no differences were demonstrated. The absolute error on the small distance when matching with the impaired limb showed similar ranks for the groups with large and the small differences in MRD (U = 22.0, z = -0.26, p = 0.8, average ranks 7.2 and 7.8 respectively). Also for the medium and the large distance no differences were found between the ‘less than 10% group’ and the ‘more than 10% group’ (Medium: U = 12.0, z = -1.55, p = 0.12, average ranks = 5.5 vs. 9.0; Large: U = 15.0, z = -1.16, p = 0.25, average ranks = 9.0 vs. 6.4).

Discussion

In order to better understand the impact of Spastic Hemiparetic Cerebral Palsy (SHCP) on position sense during bimanual tasks, the current study compared the performance of children with SHCP and TD children in a typical contralateral arm matching task. We found that children with SHCP matched the position of the reference arm less accurately than TD children as reflected in larger matching errors for both the impaired and less-impaired arm. Previously, Wann (1991) has shown similar bilateral deficits in a small group of children with mixed CP-diagnosis, i.e. quadriplegia and diplegia where the condition is caused by a lesion to the left and right hemisphere. Yet, our results demonstrate that also children with unilateral brain damage have difficulties with matching the position of the upper limbs (without visual information), which is in congruence with Lee et al. (1990) who reported similar findings in a case study with two children with SHCP. Interestingly, the performance in the current contralateral matching task appeared to depend on the range of the reaching movement required to match the target. In both the SHCP and the TD children endpoint error gradually increased as a function of the initial distance between the reference limb and the matching limb, i.e. at the start of the trial. In contrast to previous research that showed a drop in precision when localizing targets further away from the body (i.e. larger distance relative to the body; Adamo et al., 2007; van Beers et al., 1998), the distance effect found in the current study was independent of the target position relative to the body. Rather, the accuracy in this matching task was affected by the distance of the reaching movement irrespective of whether the movement was to a proximal or a distal target. It should be noted that this effect was stronger for the SHCP than for the TD children. In addition, further analysis showed that performance of the two groups only differed significantly in the medium and large distance condition.

What makes matching more prone to error when the initial distance between the effector and the target is larger? The cause of this distance effect might be related to the nature of movements children perform and practice as part of their daily routine. Daily movements in which both limbs are involved are usually movements in which the limbs are relatively close to each other, for example cutting a piece of bread, typing on the computer, or playing with a doll. As a result it is conceivable that the joint-position sense is better developed within the daily range of motion and less developed (less specific) outside that range. Furthermore, larger reaching movements are also more prone to signal-dependent noise as they require neural command signals of a greater intensity, which come with increased variance of noise (Goble, 2010; Harris & Wolpert, 1998). This phenomenon is expected to amplify the endpoint error of movements with larger amplitudes. In addition, for children with SHCP, involuntary muscle contractions associated with spasticity can lead to a situation in which the muscle tends to remain in a shortened position. This restriction in range of motion may cause length-related changes in the muscle-tendon complex and can eventually lead to a loss of joint range, or contracture (Ada, O'Dwyer, & O'Neill, 2006). Although spasticity may impede the movement required in the present study, it has to be noted that the movement was self-paced and within the range of motion of the impaired limb which should have limited the impact of the (high) velocity depended reaction. If the restriction in range of motion would explain the difference in matching accuracy between the SHCP and the TD group, more undershoot would be expected in the SCHP group (in particular for the spastic impaired arm) compared to the TD group. Yet, both groups undershot the target in the majority of the trials and there was no difference between the children with SHCP and the controls, or between the impaired and the less-impaired arm. Moreover, children with low levels of spasticity undershot the target in as many trials as the children exhibiting higher levels of spasticity and the size of the absolute error neither differed between these groups. A similar finding was demonstrated for the difference in MRD: the group with larger differences in MRD between the impaired and less-impaired arm did not show significantly more frequent undershoots or larger absolute errors than the group with smaller differences in MRD. Therefore, although we cannot exclude that the restricted range of motion in children with SHCP may have contributed to the larger endpoint errors at the large distance, the present results suggest that a compromised motor system cannot fully account for the lower matching accuracy in the SHCP group and the high prevalence of undershoot.

In addition to the diminished matching ability of the impaired arm, larger endpoint errors for the less-impaired arm compared to the dominant arm in the medium and large distance condition indicate, in agreement with previous research (Chrysagis et al., 2007; Goble, Hurvitz et al., 2009; Wingert et al., 2009), that SHCP could be associated with sensory problems in conjunction with their motor problems. The performance in the contralateral matching task is the combined result of a number of interacting factors. Afferent proprioceptive signals determine the position of the reference arm. This information is processed at cortical level leading to efferent motor commands which move the contralateral arm to the felt target position. Finally, afferent proprioceptive signals coming from the matching arm may be used to fine tune and match the position of the reference arm. It is impossible to pinpoint the origin of a matching problem on the basis of our findings, however a detailed comparison of the performance of the impaired and less-impaired limb may provide more insight into the specific difficulties encountered by children with SHCP in tasks requiring bimanual control. A first question that needs to be addressed is whether the matching difficulties may be explained by a deficit at the cortical level only. A deficiency in mapping proprioceptive signals from the reference arm onto an egocentric reference frame is likely to result in distance independent matching errors for both arms, i.e. the matching error would be the same for both arms on each distance. However, the finding that performance of the limbs of children with SHCP was only comparable (with each other and with the TD group) in the small distance condition appears to be inconsistent with this notion and suggests that deficits occur both at cortical level and at the level of the muscle. Secondly, while the impaired arm located the target less accurately than the less-impaired in the large distance condition, the opposite was found in the medium distance condition. This is in contrast with the TD children where the endpoint error was similar for both arms in all three conditions and raises the question whether position sense may be affected in the less-impaired arm of children with SHCP too. Based on purely unimanual pointing tasks, Goble et al. (2009) and Wingert et al. (2009) concluded that position sense of the less-impaired arm was not reduced. The implication would then be that the larger matching errors of the less-impaired limb for the medium distance condition in our study were caused by disturbed afferent information originating from the impaired reference limb only. This would suggest that SHCP would affect the accuracy of position sense when the impaired limb is used as a static reference (or target) more than when it is actively involved in the reaching movement. However, given the fact that involuntary spastic contractions primarily emerge when the affected muscle is stretched (i.e. dynamic rather than static conditions) the aforementioned suggestion seems to be counterintuitive. Thus while decreased position sense of the impaired limb is likely to contribute to the matching errors of the less-impaired limb, at this moment the contralateral matching task does not allow us to exclude difficulties at the level of the less-impaired arm either. At last it should also be noted that in the current study the differences between the impaired and the less-impaired side may also be related to the fact that the target locations were based on the smaller maximum reaching distance of the impaired limb. This meant that the less-impaired limb operated within smaller range of movement relative to its maximal range than the impaired limb, which may be partly responsible for the smaller error of the less-impaired limb at large distances.

To summarize, although the contralateral matching task is unable to isolate position sense deficits of the impaired and less-impaired arm, the current results demonstrate that children with SHCP are clearly disadvantaged when performing skills that involve both arms. Accurate positioning of one arm relative to the position of the other arm, which is required in numerous manual skills, is impaired regardless of which arm is used.

Finally, it has been suggested that tasks requiring processing and mapping of proprioceptive information are subserved by a fronto-parietal network that is mainly located within the right hemisphere (reflected in a left hand proprioceptive advantage for right handers; Goble & Brown, 2008). This is consistent with findings of Goble et al. (2009) demonstrating poorer proprioceptively guided matching in individuals with right hemispheric damage than in individuals with a left hemispheric damage. Reinspection of our data (6 children with right hemispheric damage vs. 8 children with left hemispheric damage) did not reveal such a difference. Since we were unable to match these two groups for size and specific location of the lesion, caution is warranted when interpreting these results. Moreover, other findings of Goble show that left-handed individuals have a right hand advantage for proprioceptive tasks (Goble, Noble, & Brown, 2009), indicating that other factors related to practice and specific function of the hand are likely to contribute to the left – right differences in position sense. Altogether without controlling for important confounding factors, such as specific location of the lesion, size of the lesion, functionality of the impaired arm etc., we believe it is premature to compare SHCP children with left and right hemispheric damage.

In conclusion, the results of the present study demonstrate that children with SCHP exhibit severe deficiencies in accurate positioning of one arm relative to the position of the other arm when compared to TD children. Despite the fact that with a contralateral matching task we cannot draw conclusions on the origin of the proprioceptive deficits, it is suggested that the unilateral proprioceptive deficits reported by previous studies, severely hamper the matching of the limbs. This deficit is particularly visible when the initial distance between the target and the matching arm is large (irrespective of target position relative to the body).

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Chapter 4

Visual feedback of the non-moving limb improves active joint-position sense of the impaired limb in Spastic Hemiparetic Cerebral Palsy

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Abstract

This study examined the active joint-position sense in children with Spastic Hemiparetic Cerebral Palsy (SHCP) and the effect of static visual feedback and static mirror visual feedback, of the non-moving limb, on the joint-position sense. Participants were asked to match the position of one upper limb with that of the contralateral limb. The task was performed in three visual conditions: without visual feedback (no vision); with visual feedback of the non-moving limb (screen); and with visual feedback of the non-moving limb and its mirror reflection (mirror). In addition to the proprioceptive measure, a functional test (Quality of Upper Extremity Skills Test [QUEST]) was performed and the amount of spasticity was determined in order to examine their relation with proprioceptive ability. Results showed that the accuracy of matching was significantly influenced by the distance that had to be covered by the matching limb; a larger distance resulted in a lower matching accuracy. Moreover it was demonstrated that static (mirror) visual feedback improved the matching accuracy. A clear relation between functionality, as measured by the QUEST, and active joint-position sense was not found. This might be explained by the availability of visual information during the performance of the QUEST. It is concluded that static visual feedback improves matching accuracy in children with SHCP and that the initial distance between the limbs is an influential factor which has to be taken into account when measuring joint-position sense.

Introduction

Cerebral palsy (CP) is, with an incidence of 2-2.5 per 1000 living births, one of the most common childhood disorders (Lin, 2003). The condition is caused by damage to the brain and/or pyramidal tract and depending on the location of the lesion and the clinical outcome of the damage, different forms of CP are distinguished. In Spastic Hemiparetic Cerebral Palsy (SHCP) the damage is limited to one side of the brain leading to impaired control of muscle tone and spasticity in the lower and upper limbs on the contra-lesional side of the body (Albright, 1996). Although SHCP is classed as a unilateral condition, recent studies have highlighted that children with SHCP have motor difficulties beyond their unilateral deficits. The spasticity of the impaired limb limits the performance of bimanual tasks and evidence suggests mild motor impairments in the unaffected limb as well (Steenbergen & Meulenbroek, 2006).

Impairments as spasticity are often accompanied by disturbances in proprioception (Cooper, Majnemer, Rosenblatt, & Birnbaum, 1995; Odding, Roebroeck, & Stam, 2006). Proprioception is a complex somatosensory modality that consists of two components: kinaesthesia and joint-position sense. Kinaesthesia is defined as the sense of limb movement whereas joint-position sense is referred to as static limb position (Goble, Lewis, Hurvitz, & Brown, 2005). Proprioception plays a major role in performing and controlling movements including updating motor plans based on e.g. monitoring movement execution through comparison of predicted and actual movement outcomes (Goble, 2006). A number of studies have demonstrated that the proprioceptive ability of children with SHCP is impaired (Goble, Hurvitz, & Brown, 2009; Wann, 1991; Wingert, Burton, Sinclair, Brunstrom, & Damiano, 2009), and there are indications that the impaired limb has a poorer proprioception than the less-impaired limb (Goble, Hurvitz et al., 2009; Wingert et al., 2009). Furthermore, in addition to the differences in proprioception between the limbs, Goble, Hurvitz et al. (2009) also found a difference in proprioceptively guided matching tasks between individuals with a left brain lesion and individuals with a right brain lesion. In individuals with a right hemispheric lesion (RHL) the proprioceptive ability was more impaired than in individuals with a left hemispheric lesion (LHL). Goble's findings can be supported by neuroimaging studies which showed that the right hemisphere is more activated during the performance of a proprioceptive task (Naito et al., 2005).

Although proprioception is impaired in individuals with SHCP, they are still able to sustain a certain level of movement accuracy, implying that visual information is used to attain this movement accuracy (van Roon, Steenbergen, & Meulenbroek, 2005). Indeed, studies by Wingert et al. (2009) and Wann (1991) on individuals with CP demonstrated that vision of the moving upper limb improved the performance on the joint-position task compared to a situation in which no visual feedback of the moving upper limb was available. However, Wann (1991) also showed that visual information of the non-moving hand did not improve movement accuracy in a joint-position sense task for individuals with bilateral CP. According to Wann (1991) this suggests that individuals with bilateral CP have difficulties encoding the visual and proprioceptive information into a common reference frame. However, the possibility that visual feedback of the non-moving limb might afford a reference frame for the proprioceptive information of the moving limb has not been investigated in individuals with hemiplegia. One of the explanations for the problems in encoding proprioceptive and visual information that Wann (1991) presents is that the cortical damage may have destroyed the neural structures that are necessary for egocentric mapping. This might indeed be the case for diplegic patients, but children with hemiplegia have a lesion in one hemisphere. It therefore might be possible that patients with hemiplegia are able to encode proprioceptive and visual information into a common reference frame. Therefore, the present study will examine the effect of visual feedback of the non-moving limb on the contralateral matching performance of the moving limb in this population. Given the asymmetry in proprioception in hemiplegia but also given the fact that only one hemisphere is damaged, it can be expected that the visual and proprioceptive information of the non-moving (less-impaired) upper limb might be integrated into one egocentric reference frame for the moving (impaired) upper limb (Jeannerod, 1986; von Hofsten & Rosblad, 1988; Wann, 1991), facilitating the contralateral matching in comparison to a situation in which no visual feedback is available.

In addition to the effect of visual information of the non-moving limb, the current study investigates the effect of mirror visual feedback of the non-moving limb on the matching accuracy during a contralateral matching task in children with SHCP. Mirror visual feedback has been demonstrated to have a positive effect on the bimanual coordination and neuromuscular activity in children with SHCP (Feltham, Ledebt, Bennett, Deconinck, Verheul, and Savelsbergh 2010; Feltham, Ledebt, Deconinck, & Savelsbergh, 2010). However, Holmes & Spence (2005) showed that manipulating the position of the moving hand (behind the mirror) influenced unimanual reaching movements in typically developed (TD) adults negatively. They suggested that this was the result of an integration of visual and proprioceptive information of the non-moving limb which caused a bias in the felt initial position of the moving hand. It can thus be hypothesized that providing mirror visual feedback of the non-moving (less-impaired) upper limb (thus seeing two non-moving upper limbs), would deteriorate the contralateral matching performance of the impaired upper limb in children with SHCP. In the forthcoming, visual feedback of the non-moving limb will be referred to as static visual feedback and visual feedback of the moving limb will be referred to as dynamic visual feedback. Mirror visual feedback of the non-moving limb will be referred to as static mirror visual feedback.

Literature on the relationship between impaired proprioception and other impairments in CP as well as the relationship with the activity level is scarce. The relationship with spasticity was assessed in the study of Chrysagis, Skordilis, Koutsouki and Evans (2007) who showed that an increase in spasticity was related to a decreased performance on an active joint-position sense task. Accordingly, Tardieu, Tardieu, Lespargot, Toby, and Bret (1984) stated that spasticity causes disturbances in the muscle spindle functioning leading to inappropriate kinaesthetic feedback (Chrysagis et al., 2007). However, the relationship between arm/hand functionality and joint-position sense has, to the best of our knowledge, not been examined yet. In order to get more insight into the influence of spasticity on joint-position sense and to clarify the impact of an impaired joint-position sense on daily functioning, the current study will investigate these two relationships.

In general, the present study aimed to get more insight into the proprioceptive impairments of the impaired and the less-impaired upper limb in children with SHCP. We assessed the role of static visual feedback and static mirror visual feedback on joint-position sense of the upper limbs using three different visual conditions: a no vision condition without any visual feedback of both limbs, a screen condition in which only the non-moving reference limb was visible (static visual feedback) and a mirror condition in which the non-moving reference limb was visible and its reflection in the mirror (static mirror visual feedback). It was hypothesized that static visual feedback of the less-impaired limb would improve the movement accuracy of the impaired limb compared to the situation without visual feedback. In addition, it was expected that static mirror visual feedback would create a conflict situation between the visual and proprioceptive feedback which would result in a deteriorated performance.

Furthermore, the current study aimed to examine the relationship between one of the main impairments in CP, spasticity, and the impaired proprioception in CP, and between the impaired proprioception and the arm/hand functionality. It was hypothesized that a higher degree of spasticity would be related to an impaired joint-position sense which would in turn be linked to a deteriorated arm/hand functionality. Finally, differences in joint-position sense impairment between left and right hemispheric brain lesions were examined. Following the findings of Goble, Hurvitz et al. (2009) it was hypothesized that individuals with a right hemispheric lesion would have a more deteriorated joint-position sense than individuals with a left hemispheric lesion.

Methods

Participants

Fourteen children with SHCP participated in the study (age 12.6 ± 1.95 years). 6 children had a right hemispheric lesion and 8 children had a left hemispheric lesion. Individual participant characteristics can be found in Table 4.1. None of the participants had any neuromuscular disorder other than SHCP, pain in either of the upper limbs, visual neglect, visual impairments not corrected to normal, mental retardation, or received a treatment with Botulinum toxin in either of the arms in the past six months preceding the measurement. The children with SHCP were recruited through the Dutch society for children with a physical handicap and their parents (BOSK). Participants' parents provided written informed consent prior to testing. All procedures were approved by the institutional research ethics committee and in accordance with the Declaration of Helsinki.

Measures of functionality

Before the actual start of the experiment different measures were performed to examine the participants' body functions. Additional information about the child's disorder was obtained from a general questionnaire, filled in by the parents, with questions about e.g. the cause and severity of the disorder and limitations the child faces in daily life. In addition, the parents were asked to fill in The Functional Independence Measure for children (WeeFIM). The WeeFIM measures the functional abilities in activities of daily life like the ability to feed, dress and bathe (Ottenbacher, Hsu, Granger, & Fiedler, 1996). For the current study only the WeeFIM motor items were used.

Grip strength was determined for each upper limb, using a hand-held dynamometer measuring the average of three maximum voluntary contractions in kilograms (JAMAR, digital hand dynamometer, Clifton, USA).

Table 4.1: Participant characteristics. For each participant the age in years, sex, side of the brain lesion, grip strength of the (less-) impaired arm, score of the Tardieu, WeeFIM, and MACS and aetiology are presented.

P	**Age (years)**	**Sex**	**Side brain lesion**[a]	**Grip strength impaired/ less-impaired limb (kg)**	**TSelbow (flex-ext)/ TSwrist (flex-ext)**[b]	**WeeFIM/ MACS**	**Aetiology**
1	13.4	M	Right	11.7/52.3	3-1/2-2	78/3	O_2 shortage during birth
2	10.5	M	Right	4.0/44.0	3-1/3-0	88/3	Thrombosis
3	10.8	M	Right	12.3/30.0	2-1/1-0	91/2	Unknown
4	14.5	M	Right	7.3/52.3	2-2/2-0	62/3	Schizen-cephaly
5	13.6	M	Right	14.7/52.0	2-2/0-0	91/2	Cerebral infraction
6	10.8	F	Right	4.7/22.0	2-1/0-0	52/3	Cerebral Haemorrhage
7	12.1	F	Left	2.0/63.7	2-0/2-1	91/3	Thalamus infarction at birth
8	15.5	M	Left	60.3/105.7	2-0/0-0	76/1	Unknown
9	9.3	M	Left	23.3/49.7	2-0/0-0	91/1	Cerebral infarction
10	13.1	F	Left	25.0/69.7	2-2/0-0	91/2	Cerebral infarction
11	14.4	M	Left	0.0/104.0	2-0/0-0	81/2	Cerebral haemorrhage
12	12.5	M	Left	0.0/62.0	2-2/2-0	59/3	Cerebral infarction
13	14.3	M	Left	13.6/101.3	2-2/1-0	71/3	Unknown
14	10.6	M	Left	24.7/69.0	0-1/0-0	87/2	O_2 shortage during birth

[a] The impaired arm is the arm contralateral to the brain lesion.
[b] TS = Tardieu Score of the impaired limb. (flex/ext) are separate scores for flexion and extension.

The Quality of Upper Extremity Skills Test (QUEST; DeMatteo et al., 1992) was performed to qualify the functional ability of the arms and hands of each participant. This test consists of 7 domains, however for this study only the parts about “Dissociated movements” (part A) and “Grasps” (part B) were conducted since these two domains were specifically related to the task the children had to perform during the measurement. The QUEST is validated for children between 18 months and 8 years of age (DeMatteo et al., 1992). However, although the mean age of our population is 12.6 years it was still chosen to use the QUEST since this test is more extensive than other tests that measure the functioning of the upper limbs. Based on the items of the two included parts of the QUEST and the related scoring criteria we calculated separate scores for the impaired and the less-impaired limb. A higher score on this selection of QUEST items represents a better functionality. Table 4.2 presents the individual QUEST scores. The performances of the QUEST were recorded with a digital video camera (JVC Hard disk Camcorder, HDD F1.2, GZMG40E) in order to score the performances afterwards. Two experimenters analyzed the video tapes independently. The inter-rater reliability was high ($r = 0.92$, $p < 0.001$).

In addition to the QUEST, the Manual Ability Classification System (MACS) level was determined. The MACS describes how children use their hands during object handling and their need for assistance to perform manual skills in everyday life (Carnahan, Arner, & Hagglund, 2007). The severity of performance limitation and the degree of required assistance increases for each MACS level from 1 to 5. The MACS levels and their specifications are depicted in Table 4.3.

The degree of spasticity was determined by a qualified physiotherapist using the Tardieu Scale. The assessment involved passive movement of the arm in the sagittal plane, first as slow as possible and second as fast as possible, while the child was seated on a chair with the knees bend in 90°. The physiotherapist quantified the spasticity of the arm muscles (Biceps Brachii Brevis, Triceps Brachii Longus, flexors and extensors of the wrist) during the fast velocity stretch according to the criteria of muscle reaction for grades 0-3. The definition of each grade is depicted in Table 4.4. The Tardieu score averaged for the Biceps and the Triceps was further used for analysis.

Table 4.2: QUEST scores; Total score and scores of Part A (dissociated movements) and Part B (grasps) for each limb.

P	**Total score**	**Part A impaired limb**	**Part A less-impaired limb**	**Part B impaired limb**	**Part B less-impaired limb**
1	72.2	60.0	99.2	86.7	100
2	51.1	57.0	100	50.0	80.0
3	82.5	86.6	99.1	81.7	88.3
4	65.3	72.5	100	60.0	80.0
5	68.5	66.5	100	73.3	90.0
6	52.6	64.8	99.2	48.3	85.0
7	77.4	71.5	100	85.0	100
8	96.4	98.4	100	96.7	98.3
9	95.9	99.2	100	93.3	93.3
10	81.7	78.1	100	86.7	100
11	55.2	54.8	100	60.0	100
12	51.4	54.7	100	55.0	93.3
13	63.0	70.7	98.4	65.0	95.0
14	85.1	77.3	98.4	95.0	95.0

Table 4.3: Description for each MACS level.

MACS level	Description
1	Handles objects easily and successfully.
2	Handles most objects but with somewhat reduced quality or speed of achievement.
3	Handles objects with difficulty; needs help to prepare or modify activities.
4	Handles a limited selection of easily managed objects in adapted situations.
5	Does not handle objects and has severely limited ability to perform even simple actions.

Table 4.4: Tardieu scale scoring the quality of muscle reaction to stretch.

0	No catch, no resistance.
1	Light resistance without clear catch.
2	Clear catch followed by a release.
3	Clear catch, no release.

Procedures

The child was seated on a height adjustable chair at a height adjustable table with the knees 90° flexed. Joint-position sense was assessed using a custom made device consisting of two handles, each on a separate track fixed to a horizontal panel. The tracks were 20 cm apart, parallel to each other, and perpendicular to the medio-lateral axis of the trunk. The handles could be moved within a range of 56 cm. The children were positioned such that the centre of the body was located in between the two tracks, and with the beginning of the track at 15 cm from the upper body. The position of the handles was recorded outside the wooden device using one Optotrak unit with three infrared cameras (3020 Optotrak, Northern Digital Inc., Waterloo, Canada). The experimental setup is depicted in Figure 4.1.

Before the start of the measurement, the maximum reaching distance of the impaired arm was determined (MRD) in order to scale the different matching positions across subjects. MRD was the distance from the start of the track to the position of the handles when the elbows were extended as far as possible without bending the trunk forward. If a participant was unable to grip the handle due to physical impairment, the experimenter placed the hand on top of the handle. All participants were able to hold the handles during the whole experiment.

The active joint-position sense task required participants to match the position of one limb (reference limb), fixed at 25%, 50%, or 75% of the MRD, by actively moving the other limb (matching limb). The task was performed with either the less-impaired limb or the impaired limb and the matching started at the MRD (distal) or at the beginning of the track (proximal). The matching task was performed in three different visual conditions: a no vision condition (both hands were not visible), a screen condition (only the reference hand was visible), and a mirror condition (only the reference hand was visible and its reflection in the mirror). The position of the reference limb (3), the matching limb (2), the start position of the matching limb (2), and the visual conditions (3) resulted in 36 trials. The conditions were randomly presented to the participant but all trials with the same matching limb were kept together even as the trials within one visual condition. Prior to data collection 3 practice trials were conducted to familiarize the participant with the test setup. In order to keep the children motivated they were told that the better their performance the more points they could get. At the end of the experiment they could trade their points for a small gift.

Figure 4.1: Experimental setup during the no vision (left panel), screen (middle panel), and mirror (right panel) condition.

Data analysis

A custom made Matlab program (The Mathworks, Inc.) was used to determine the absolute difference (error) between the position of the reference limb and the position of the matching limb at the end of the movement. The end of the movement was indicated by visual inspection (see Figure 4.2).

Goble, Coxon, Wenderoth, Van Impe, & Swinnen (2009) stated that several studies that measured proprioceptive acuity found larger errors for the matching of targets farther from the body in contrast to targets closer to the body. However, in these studies the starting position was the same for all trials and hence it can be argued that the distance that has to be covered by the matching limb is the influencing factor instead of the position relative to the body. This idea is supported by Smorenburg, Ledebt, Deconinck, & Savelsbergh (2012) who found larger errors when the distance covered by the matching limb was larger. Therefore the current study combined the two starting positions (distal, proximal) of the matching limb and the three positions of the reference limb (25%, 50%, 75% of the MRD) into three distances that had to be covered by the matching limb (small, medium, large).

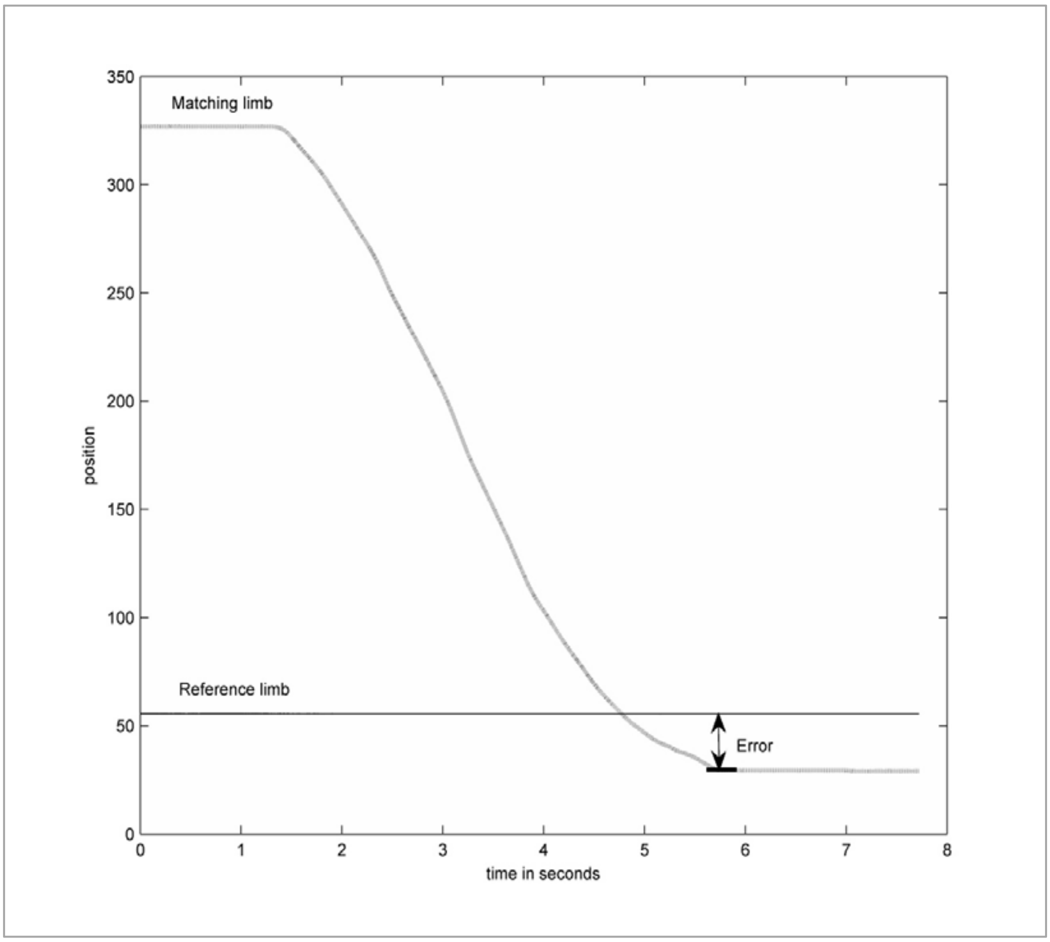

Figure 4.2: Example of a movement pattern. The arrow indicates the distance between the limbs at the end of the movement.

Statistical analysis

A repeated measurement ANOVA was performed with Distance (small, medium, large), Matching limb (impaired, less-impaired) and Visual condition (mirror, screen, no vision) as within factors. Lesion side (left hemispheric lesion [LHL], right hemispheric lesion [RHL]) was taken as between factor. If the sphericity assumption was violated, Greenhouse Geisser adjustments were made. Post hoc comparisons for the interaction effects were performed with the Fishers' LSD test.

Correlations

Correlations were calculated using the Pearson's correlation coefficient (r). For the correlations with the Tardieu Scale, Spearman's correlation coefficient was used (r_S).

Results

Matching accuracy

The accuracy of active matching was significantly influenced by Distance ($F_{(1.2,\,14.1)} = 8.71$, $p = 0.008$), showing a general trend that the absolute error became gradually larger with larger matching distances. Other main effects were absent, but all factors were involved in second order interactions (Hand x Distance: $F_{(2,24)} = 3.99$, $p = 0.032$; Visual condition x Distance: $F_{(4,48)} = 3.81$, $p = 0.009$) and a third order interaction (Hand x Distance x Visual condition: $F_{(4,48)} = 3.26$, $p = 0.019$; see Figure 4.3). Figure 4.3 reveals similar trends for all visual conditions in the less-impaired limb and the screen and mirror conditions in the impaired limb. In accordance with the main Distance-effect smaller errors were made in the small distance condition, except for matching with the less-impaired limb in the mirror condition where no significant differences between distances were found. The differences between the two limbs and between the visual conditions were related to the deviant profile of the no vision condition for the impaired hand. Matching large distances with the impaired limb without visual information resulted in significantly larger errors than in the mirror or screen condition. In addition, the impaired limb showed a similar or larger error as the less-impaired limb with exception of the medium matching distance in the no vision condition. Matching with the impaired limb in this condition (medium, no vision) yielded smaller errors than for the less-impaired limb, whereas the latter was more accurate than the impaired limb in the large distance, no vision condition. Finally, no differences in accuracy of active matching were found between LHL and RHL.

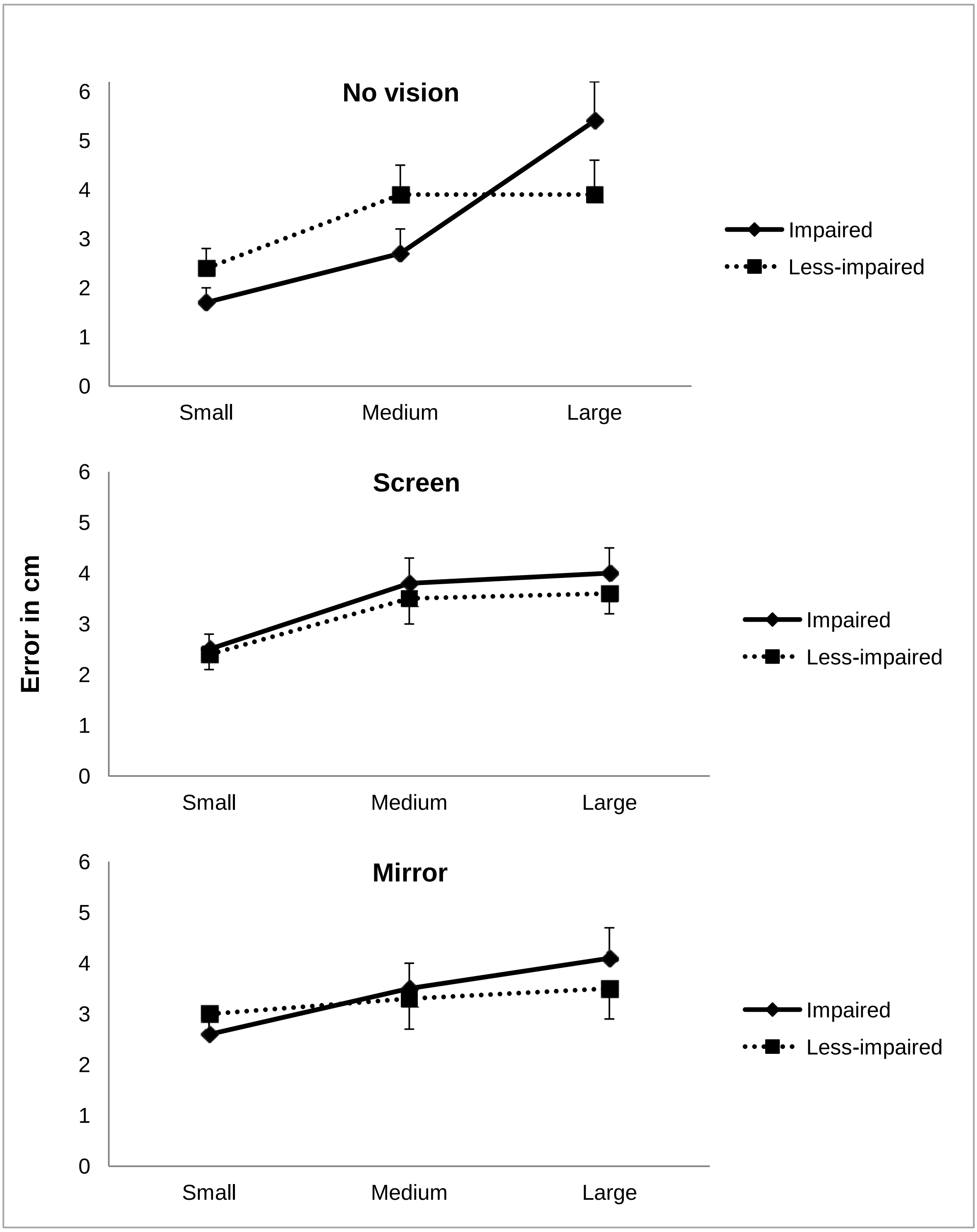

Figure 4.3: Absolute error (in cm) in the three visual conditions (no vision, screen, mirror) for the impaired (solid line) and the less-impaired arm (dashed line) on the three distances (small, medium, large).

Functionality (QUEST) and Spasticity

QUEST vs. active joint-position sense

A significant negative correlation was revealed between the QUEST part A (dissociated movements) of the impaired limb and the error on the active joint-position sense task of the impaired limb in the screen condition for the large distance (r = -0.70, p = 0.006).

QUEST for left- and right hemispheric lesions

The QUEST score part A (dissociated movements) and the QUEST score part B (grasps) of the impaired upper limbs were not significantly different between the LHL and the RHL group. Moreover, for the less-impaired limb no difference between the two groups was revealed for the QUEST score part A, but for the QUEST score part B the RHL group had a higher score than the LHL group (mean difference = 9.65, p = 0.006).

Spasticity vs. active joint-position sense

A significant correlation between the mean Tardieu score of the Biceps and the Triceps and the absolute error on the active task was found. A higher Tardieu score was related to a smaller error of the impaired limb in the no vision condition for the large distance (r_s = -0.54, p = 0.047). This relation is depicted in Figure 4.4.

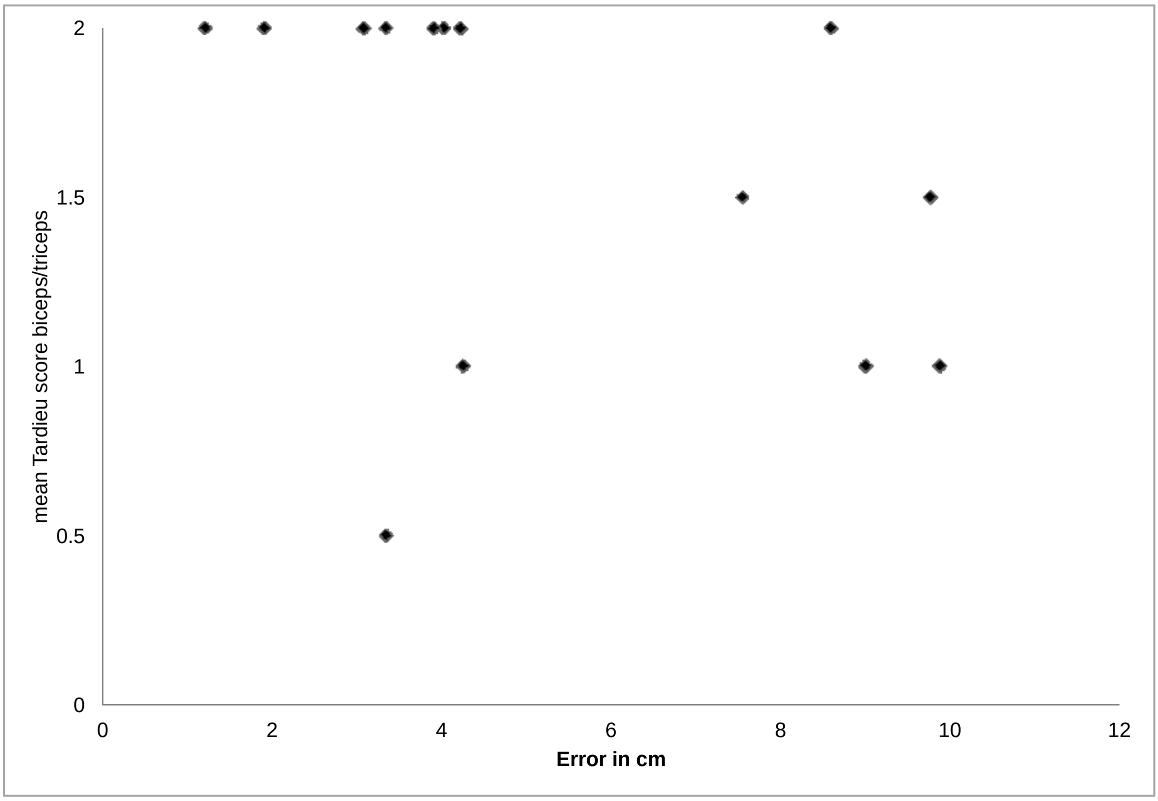

Figure 4.4: Correlation between the Tardieu score averaged for the Biceps and Triceps and the error on the active joint-position sense task of the impaired limb in the no vision condition for the large distance.

Discussion

The current study aimed to get more insight into the integrity of proprioception in the impaired and less-impaired limb in children with SHCP. In an active joint-position sense task, different visual conditions were used in order to investigate the effect of static visual feedback and static mirror visual feedback on joint-position sense. In addition, the relation between joint-position sense and spasticity and joint-position sense and arm/hand functionality was investigated. Finally, following the findings of Goble, Hurvitz et al. (2009) we examined differences in joint-position sense between individuals with a right hemispheric lesion and individuals with a left hemispheric lesion.

A general finding in this study was that the position of the reference limb could be matched with greater accuracy when the distance to be covered was smaller, irrespective of which limb was used to match and irrespective of the initial position of the reference limb (in the proximity of the body or further away). This finding is in agreement with previous results in typically developing children (Goble & Brown, 2008; Goble, Lewis, & Brown, 2006) and children with SHCP (Smorenburg et al., 2012) A physiological phenomenon that may explain the larger absolute errors for longer reaching or matching distances is the signal-dependent noise on a motor command. According to this principle the variance of the noise on a neural control signal increases with the size of the signal (Harris & Wolpert, 1998). This would suggest that for larger distances, requiring the generation of a larger command signal, the variance of noise becomes larger, which will hamper the accurate matching of the upper limbs. In addition to this physiological explanation, it is assumed that factors associated with daily functioning may play a role in the distance-effect, especially when considering the matching task used in the current study. Goble et al. (2005) suggested that the improvements in the acuity of joint-position sense when comparing children and adolescents are partly the result of experience-driven processes. Our daily movement repertoire is diverse, but with respect to grasping and reaching movements the range of motion is typically kept relatively small, which may lead to a distance-specific specialization of proprioception. In this respect it is interesting to note that in the current experiment the error score was highest when matching large distances with the impaired arm. Due to the spasticity, which tends to shorten the muscles leading to partial immobility of this arm (Love et al., 2001), children with SHCP might avoid using the arm for tasks involving larger ranges of motion. This substantial increase in absolute error for the large distance condition was absent when matching with the less-impaired arm. Although a better acuity of this less-affected arm can be expected, this finding is still remarkable because the contralateral matching task involves the utilization of afferent proprioceptive information from both the reference (impaired) and the matching (less-impaired) arm.

Comparison of the error score across visual conditions indicates that static visual feedback of the reference limb has the capacity to improve joint-position sense, in particular when matching large distances with the impaired arm. This finding is in contrast to those of Wann (1991) who found that a group of children with mixed diagnoses of CP did not benefit from visual information of the reference limb and target in a similar matching task. Wann (1991) showed that the performance of the children with CP for tasks requiring crossmodal matching (between sensory modalities, i.e. vision and proprioception), was lower than in all other conditions where intramodal matching was possible (within one sensory modality). It was concluded that CP was associated with a reduced ability to generate an egocentric frame of reference needed for accurate mapping between sensory modalities. It is important to note that the children participating in Wann's study all had suffered bilateral damage to the brain (diplegia and quadriplegia). Our results then imply that in children with unilateral damage to the brain, crossmodal mapping is not disturbed to a similar extent as in diplegic and quadriplegic patients, and still allows the encoding of sensory signals into a common egocentric frame of reference. The beneficial effect of vision in a situation where spasticity compromises matching acuity most (large distance matching with impaired hand), suggests that joint-position sense in children with SHCP seems to be affected by a distortion of the physiological function of the somatosensory organs, rather than by a deficit in higher sensory motor function. Our finding that static visual feedback of the less-impaired limb improves the matching accuracy might potentially be interesting for therapeutic interventions in order to improve the joint-position sense of the impaired limb. If training with static visual feedback of the less-impaired limb can improve the joint-position sense of the impaired limb, this might have implications for the daily functioning of the children. The focus nowadays is primarily on improving motor behaviour by practicing, but since proprioception is an important factor in movement control, this might be another angle of approach in order to improve daily functioning in children with SHCP.

Despite the beneficial effects of static visual feedback, no detrimental effects of static mirror visual feedback were found. Based on the findings of Holmes and Spence (2005) it was expected that static mirror visual feedback would deteriorate the matching accuracy, especially of the impaired limb. However, Holmes and Spence (2005) showed also that a longer exposure time to the mirror resulted in larger errors. The short exposure time in the current study might explain why we did not find an effect of the mirror in the active joint-position sense task. Moreover, in general, proprioceptive information is more reliable under active than under passive conditions. It can be expected that perceived hand position will be less affected by (discrepant) mirror visual feedback in an active compared to a passive condition (Chokron, Colliot, Atzeni, Bartolomeo, & Ohlmann, 2004; Holmes & Spence, 2005; Van Beers, Wolpert, & Haggard, 2002). It is therefore suggested to examine the differences in mirror effect between an active and a passive joint-position sense task.

Based on the study of Goble, Hurvitz et al. (2009) we expected that differences in joint-position sense between the upper limbs and the effects of visual information would be different for individuals with a left hemispheric lesion and individuals with a right hemispheric lesion, but in the present study no effect of lesion side was found. Differences in task (ipsilateral remembered vs. contralateral matching) between our study and the study of Goble, Hurvitz et al. (2009) might have caused these discrepant findings. Moreover, in both studies no specific information about the location of the brain lesion is present which makes it difficult to draw clear conclusions. However, the current study examined the functional level of the participants by means of the QUEST, which might shed a light on the severity of the condition. It was shown that participants with a LHL and participants with a RHL had the same mean QUEST scores for the impaired side of the body. Although both groups in the study of Goble, Hurvitz et al. (2009) had similar spasticity scores, no information about the functional level was available. Without this information it is impossible to determine whether differences in joint-position sense between individuals with LHL and RHL are actually caused by the side of the lesion or by other factors related to the severity of the condition.

Finally, we looked at the relation between spasticity and joint-position sense and between arm/hand functionality and joint-position sense. One significant correlation between spasticity and joint-position sense was found. However, a close look on the significant correlation shows that seven individuals with a mean Tardieu score of 2 had a relative small error. The other seven participants showed a more scattered distribution. Hence it can be argued that this is not a clear-cut relationship. It is possible that the participants adapted their movement velocity in order to minimize the effect of their spasticity. Since the Tardieu scale is determined at a (fast) speed by the physiotherapist, it is plausible that this speed does not match with the movement speed during the active task. The current findings are in contrast with the findings of Chrysagis et al. (2007) who found that a higher degree of spasticity was related to a more deteriorated joint-position sense. However, Chrysagis et al. (2007) used the Modified Ashworth Scale (MAS) to determine the degree of spasticity whereas we used the Tardieu scale. Although both scales are frequently used as clinical measure, the inter-rater reliability and test-retest reliability are better for the Tardieu than for the MAS (Fosang, Galea, McCoy, Reddihough, & Story, 2003; Mehrholtz et al., 2005). Nevertheless, the question remains, irrespective of the scale used, whether such clinical measures are suitable to use in studies like the current study where the participants were free to move at their own pace. We therefore suggest that the relationship between proprioception measured with self-paced movement and the level of spasticity (measured with the Tardieu or the MAS) should take into account both the velocity of the self induced movement and the velocity of the passive movement used to evaluate spasticity.

Correlations between the arm/hand functionality and joint-position sense revealed that a higher QUEST score was related to a higher accuracy on the active joint-position sense task. However, this was only found for the QUEST score part A (dissociated movements) in relation with the accuracy of the impaired limb in the screen condition for the large matching distance. A possible explanation for the small amount of correlations between the QUEST and the active joint-position sense might be that the QUEST is performed under full vision. The visual information could compensate for the deteriorated joint-position sense whereas in the active joint-position sense task used in this study, no full compensation could take place since no visual feedback of the moving limb was available. Therefore, the absence of a significant relationship might indicate that on average the participants were able to compensate for the impaired proprioception with online visual control.

In sum, it can be concluded that static visual feedback of the less-impaired limb improved the active joint-position sense of the impaired limb in children with SHCP. Static mirror visual feedback did not have a detrimental effect on active joint-position sense. In addition, it was demonstrated that the distance that had to be covered by the matching limb had an influence on the differences between the limbs and the differences between the visual conditions. In general the error became smaller with a smaller matching distance. The relationship between matching accuracy and arm/hand functionality and matching accuracy and spasticity remains indecisive.

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Chapter 5

Matching accuracy in hemiparetic cerebral palsy during unimanual and bimanual movements with (mirror) visual feedback

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Abstract

In the present study participants with Spastic Hemiparetic Cerebral Palsy (SHCP) were asked to match the position of a target either with the impaired arm only (unimanual condition) or with both arms at the same time (bimanual condition). The target was placed at 4 different locations scaled to the individual maximum reaching distance. To test the effect of mirror visual feedback of the less-impaired arm on the matching accuracy, an opaque screen or a mirror was placed in between the arms which masked vision of the impaired arm. Absolute endpoint error was smaller in the bimanual condition compared to the unimanual condition, but there was no effect of mirror visual feedback. Inspection of the individual data, however, showed that 13 out of 23 participants did experience a positive effect of mirror visual feedback. A positive correlation between the baseline error (screen) and the improvement in accuracy with mirror visual feedback seems to suggest that individuals with lower proprioceptive accuracy in the baseline condition may benefit more from mirror visual feedback. Together these findings indicate that bimanual therapy and therapy with mirror visual feedback might be valuable approaches for rehabilitation for a subset of the individuals with SHCP.

Introduction

Cerebral palsy (CP) is the most common pediatric physical disability (Stanley, Blair, & Alberman, 2000). The condition comprises a group of permanent disorders of movement and posture due to a lesion in the foetal or infant brain. In children with spastic hemiparetic cerebral palsy (SHCP), the motor impairments are mainly lateralized (i.e. one-sided) and the upper limb is usually more affected than the lower limb (Charles & Gordon, 2006; Humphreys, Whiting, & Pham, 2000). The brain damage in SHCP might also include areas that are involved in bimanual coordination such as the supplementary motor area (SMA) and areas in the parietal lobe (Serrien, Nirkko, Lovblad, & Wiesendanger, 2001; Serrien, Strens, Oliviero, & Brown, 2002; Steyvers et al., 2003). For this reason and because many daily activities require both hands, SHCP is often found to have a detrimental effect on bimanual tasks, and hence on many tasks of daily living (Gordon, 2011; Gordon & Steenbergen, 2008; Hung, Charles, & Gordon, 2004). Yet in tasks that typically require bimanual coordination using the non-dominant (impaired) hand is avoided and while they may become adept at using this compensatory strategy, this behaviour is considered to be inefficient and slow (Charles & Gordon, 2006; Gordon & Steenbergen, 2008). Interestingly though, there is evidence to suggest that the kinematics of the impaired arm are improved when the contralateral (less-impaired) arm performs an identical (symmetrical) action (Sugden & Utley, 1995; Utley & Sugden, 1998). These studies have mainly focused on kinematic variables (e.g. speed, trajectory or timing of the two limbs) and it remains to be determined whether accuracy of matching (of the impaired arm) is also favoured in a bimanual (symmetrical) condition. This will be the focus of our study.

Steenbergen, Hulstijn, de Vries and Berger (1996) studied the arm kinematics of young adolescents with SHCP during a reach-grasp-placement task. The participants were asked to pick up a ball and place it into a hole as quickly as possible with either one hand (one ball) or with two hands (two balls). It was found that the large differences in reaction time and total movement time between the hands in the unimanual condition decreased under bimanual conditions, indicating a tendency to move the impaired and less-impaired arm and hand in a symmetrical manner (interlimb coupling). Note though, that in this study the coupling was mainly unidirectional, i.e. the result of adaptations of the less-impaired hand to the movement of the impaired hand. Using similar reaching and grasping tasks Utley and Sugden (1998) further found that coupling (temporal and to a lesser extent also spatial) happened predominantly in the first part of the movement (and not in the grasping phase) and was facilitated when movements were performed under speeded conditions. However, in contrast to the findings of Steenbergen et al. (1996) the coupling was not unidirectional, i.e. temporal synchrony was the result of adaptations in both hands (see also Sugden & Utley, 1995). Finally, Volman (2005) demonstrated that interlimb coupling in children with SHCP is not just restricted to timing of the movement but also extends to spatial features. When children with hemiplegia were asked to draw a line with one hand and a circle with the other hand, the lines became more circular and the circles became more linear compared to a single handed condition. Neither the impaired nor the less-impaired arm dominated the coupling. Taken together, these findings demonstrate that even in individuals that have suffered unilateral brain damage that led to SHCP, typical bilateral neural interactions facilitating interlimb coupling seem to be present. This coupling appears to be dependent on a number of factors such as speed and the nature of the movement. It is however not known whether this coupling influences the accuracy of a matching action. Therefore, the first question that this study will address is: Is the accuracy of matching with the impaired arm better when the less-impaired arm is moving towards the target simultaneously than when moving in isolation?

Matching accuracy can serve as a measure of proprioceptive accuracy, the sense of body parts in space, which is essential for movement performance. A previous study by Smorenburg, Ledebt, Deconinck and Savelsbergh (2012) has shown that children with SHCP perform poorer than their typically developing peers in a task where the position of one arm has to be matched with the other arm, which is indicative of deteriorated proprioceptive accuracy. If simultaneous movement of the less-impaired arm towards a target would improve the accuracy when matching with the impaired arm, this would support the integration of symmetric bimanual tasks in the training of impaired arm function.

A second phenomenon that has received a lot of attention with respect to the treatment of unilateral movement and pain disorders is mirror visual feedback (see Ramachandran & Altschuler, 2009 for a review). It is generated by placing a mirror between the upper limbs in the sagittal plane, so that one sees the real less- (or non-) impaired arm and its mirror reflection, which now is superimposed on the impaired arm. This creates the illusion of two hands moving in perfect symmetry. Mirror visual feedback has been demonstrated to alleviate (phantom) pain (McCabe et al., 2003; Ramachandran & Rogers-Ramachandran, 1996) and to improve movement performance in individuals with hemiparetic stroke (e.g. Altschuler et al., 1999; Stevens & Stoykov, 2003; Yavuzer et al., 2008). In addition, Feltham, Ledebt, Bennett, Deconinck, Verheul and Savelsbergh (2010) suggested that mirror visual feedback might be a feasible therapeutic tool for children with SHCP. Performing a bimanual inward symmetrical movement with mirror visual feedback of the less-impaired arm decreased the variability of the interlimb coupling compared to a situation in which only the less-impaired arm was visible. Furthermore, in a subsequent study the authors showed that mirror visual feedback had favourable effects on the neuromuscular activity during a symmetric bimanual movement (Feltham, Ledebt, Deconinck, & Savelsbergh, 2010). The suggestions of Feltham and colleagues were supported by a recently published study showing that 3 weeks of mirror therapy in children with SHCP resulted in improved grasp strength and upper limb dynamic position (Gygax, Schneider, & Newman, 2011). Smorenburg, Ledebt, Deconinck and Savelsbergh (2011), on the other hand, found that mirror visual feedback of the less-impaired arm did not influence endpoint accuracy of the impaired arm during unimanual matching. In this task the individuals were instructed to move the impaired limb to the position of the less-impaired limb, which was held passively at a target. In contrast to Feltham, Ledebt, Bennett et al. (2010), Feltham, Ledebt, Deconinck et al. (2010) and Gygax et al. (2011) mirror visual feedback in the Smorenburg et al. study (2011) was ‘static’, i.e. the less-impaired arm was held at the target. This discrepancy in findings seems to suggest that mirror visual feedback might only be effective when both arms are intending to move symmetrically, which is a pertinent issue that needs to be clarified before therapy with mirror visual feedback can actually be integrated in the treatment of SHCP. Therefore, the current study will examine if mirror visual feedback might have a positive effect on the endpoint accuracy of a matching task (a measure of proprioceptive acuity) when the less-impaired arm is moving simultaneously with the impaired arm (symmetric bimanual movement), and thus when the mirror visual feedback is dynamic.

Methods

Participants

Twenty five individuals with SHCP took part in the study, but 23 participants were included for analysis (14.2 ± 2.9 years, 5 females). All participants were recruited through the Dutch society for people with a physical handicap and their parents (BOSK) and the Werkenrode school in Groesbeek (The Netherlands), a special education school. Two participants were not included for analysis; one participant was not able to finish the experiment due to fatigue, and another participant had absolute error values that were more than 2 standard deviations of the mean. The participants did not have a visual impairment (which was not corrected to normal), hearing impairment, pain in either of the upper limbs, visual neglect, Botox treatment in the past six months preceding the measurement, or any other neuromuscular disorder than SHCP. Moreover, participants were required to understand basic instructions in order to perform the measurement. Table 5.1 represents the participant characteristics. For each participant the level of spasticity was determined with the Tardieu scale which ranges from 0 to 3, with a higher score indicating higher levels of spasticity. Individual scores were obtained for the Biceps Brachii Brevis and Triceps Brachii Longus and combined into one total score. Functional independence in daily life, taking into account caregiver assistance and the use of special equipment, was measured with the motor items of the Functional Independence Measure for children (WeeFIM). The participant's parents filled in the WeeFIM questionnaire. WeeFIM scores can range from 13 to 91, with a higher score representing a better functional independence. Finally, the Manual Ability Classification System (MACS) describes how children use their hands during object handling and the degree of required assistance (Eliasson, et al., 2006). The severity of performance and the degree of required assistance increases from MACS level 1 to 4. For more detailed information about the Tardieu, WeeFIM and MACS we refer to the Appendix. Prior to testing, the participant's parents provided written informed consent. All procedures were approved by the institutional research ethics committee and in accordance with the Declaration of Helsinki.

Materials and procedures

The participant was seated on a height adjustable chair at a height adjustable table with the knees flexed to 90°. On the table a custom made wooden construction was placed which consisted of two handles on two separate parallel tracks 20 cm apart (see Figure 5.1). The participant grasped the two handles (one in each hand), which could be moved in the anterior-posterior direction. The children were positioned such that the centre of the body was located in between the two tracks, with the beginning of the track 15 cm from the trunk. The position of the handles was recorded outside the wooden construction using one Optotrak unit with three infrared cameras (3020 Optotrak, Northern Digital Inc., Waterloo, Canada) at a sample rate of 200 Hz. A mirror or opaque screen, which was placed in between the tracks and perpendicular to the chest, served to elicit mirror visual feedback of the less-impaired arm or visual feedback of the less-impaired arm only.

Before the start of the measurement, the maximum reaching distance was determined (MRD). The child was asked to grasp the handles and extent the elbows as far a possible without bending the trunk forward. The MRD of the impaired arm was used to calculate the different target positions for the matching task. If a participant was unable to grip the handle due to physical impairment, the experimenter placed the hand on top of the handle. Each participant performed two tasks: a unimanual matching task and a bimanual matching task. The order of the tasks was randomly assigned to the participants. In the following paragraphs the procedures for the unimanual and the bimanual matching task will be explained.

Table 5.1: Participant characteristics. For each participant (P) the age in years, sex and impaired arm are indicated. In addition, the Tardieu scale for spasticity, the WeeFIM score and MACS level are mentioned. In the last two columns the aethiology of the disorder and the maximum reaching distances (MRD) of the impaired and less-impaired arm are given.

P	**Age (years)**	**Sex**	**Impaired arm**	**TS**[a]	**WeeFIM/ MACS**	**Aetiology**	**MRD I/LI (cm)**[b]
1	11.1	M	Left	1.5	91/2	Unknown	35.5/38
2	14.8	M	Left	2	62/3	Schizencephaly right	33/36
3	13.7	M	Left	2	78/3	O_2 shortage during birth	33/40
4	14.0	M	Left	2	91/2	Cerebral infarction	31.5/33.7
5	13.3	M	Left	2	70/2	Unknown (twins)	29/32
6	13.8	F	Left	1.5	91/2	O_2 shortage (twins)	27.3/29.5
7	13.0	M	Left	1	91/2	Hydrocephalus	20/24
8	14.5	M	Left	1	91/2	Stroke	30/31
9	14.6	M	Left	1	59/3	Streptococcen infection at 5 weeks	24/40
10	17.8	M	Left	1.5	90/1	Cerebral infarction	38/39
11	17.0	M	Left	1	91/1	Unknown	25/29
12	18.7	M	Left	0.5	88/2	Cerebral infarction	25.5/29
13	9.6	M	Right	1	91/1	Cerebral infarction	34.5/35.5
14	14.7	M	Right	2	71/3	Unknown	33/38
15	12.8	M	Right	2	59/3	Cerebral infarction	26.5/38
16	9.3	F	Right	2	85/2	Hydrocephalus	30/33.3
17	16.2	M	Right	2	76/1	Unknown	40/40
18	12.7	F	Right	1	91/3	Thalamus infarction at birth	30/32.5
19	18.7	M	Right	1.5	91/3	Cerebral infarction	33/39
20	7.9	F	Right	1	91/1	Feverish convulsion	25/26
21	17.2	M	Right	Unknown	89/3	Cerebral infarction	22/28.5
22	17.7	F	Right	1.5	91/2	Stroke	22/29
23	14.5	M	Right	0.5	91/2	Unknown	25/27

[a] TS = Tardieu scale for spasticity; mean of the individual scores for the Biceps and the Triceps.

[b] MRD = maximum reaching distance in cm for the impaired (I) and the less-impaired arm (LI).

Unimanual matching task

In the unimanual mathing task, a target was placed at 25%, 50%, 65%, or 80% of the MRD on the side of the less-impaired hand. The less-impaired hand was placed on the lap and the impaired hand was holding the handle on the other side of the mirror/screen and was not visible. The participant was asked to match the position of the target by actively moving the impaired arm (the impaired hand always started proximal to the body at the start of the track, i.e. 0%MRD). The task was performed in two different visual conditions:

a screen condition in which only the target was visible and a mirror condition in which the target and its mirror reflection were visible. Each combination of visual condition (2) and target position (4) was performed twice, which resulted in 16 trials. The order of the visual condition and the target positions were randomly assigned to the participants.

Bimanual matching task

In the bimanual matching task a target was placed at 25%, 50%, 65%, or 80% of the MRD on the side of the less-impaired arm. The participant was asked to match the target position with both hands, i.e. to move both hands towards the target as symmetrically as possible starting with the handles at the beginning of the track, i.e. 0%MRD. Similar to the unimanual task, the bimanual task was performed in two different visual conditions: a screen condition in which the target and the (moving) less-impaired arm could be seen and a mirror condition in which the participant saw the target, the (moving) less-impaired arm and its mirror reflection. Each combination of visual condition (2) and target position (4) was performed twice (16 trials in total) and the order of the visual condition and the target positions were randomly assigned to the participants.

Data analysis

Custom-made Matlab programs (The Mathworks, version 7.1) were used to analyze the kinematics and matching accuracy (absolute error) of the movement. The start of the movement was defined as the moment at which the movement velocity rose above 5 mm/s for the first time and the hand was moving in a forward direction. The end of the movement was defined as the moment at which the velocity finally fell below 5 mm/s (van Roon, Steenbergen, & Meulenbroek, 2005). Absolute error was determined as the difference in cm between the target and the impaired arm at the end of the movement. In addition, we calculated average movement velocity (cm/sec; total distance covered divided by total movement time) and relative movement smoothness. Relative movement smoothness was defined as the number of peaks in the velocity plot of the entire movement divided by the total distance covered during each movement. The number of peaks was determined by searching the velocity curve for local minima and maxima. An increase in velocity between an adjacent minimum and maximum that exceeded the threshold value (10% of the maximum velocity) was counted as a peak (Chang, Wu, Wu, & Su, 2005; Kamper, McKenna-Cole, Kahn, & Reinkensmeyer, 2002; Ledebt, Smorenburg, & Savelsbergh, in preparation).

Statistical analysis

In order to examine differences in absolute error, mean velocity and movement smoothness of the impaired arm between the unimanual and bimanual task and to examine the effects of visual feedback and target distance on these variables, a 3-way ANOVA was performed with repeated measures on the factors Task (unimanual, bimanual), Visual condition (mirror, screen), and Distance (25%, 50%, 65%, 80%MRD).

In addition, for the bimanual task differences in kinematics between the impaired and the less-impaired arm and the effect of Visual condition and Distance were investigated with a 3-way repeated measures ANOVA with Arm (impaired, less-impaired), Visual condition (mirror, screen), and Distance (25%, 50%, 65%, 80%MRD) as within factors.

The significance level was set at 0.05. In case sphericity assumptions were violated, Greenhouse-Geisser adjustments were made. Post hoc comparisons were performed with the Fishers' LSD test.

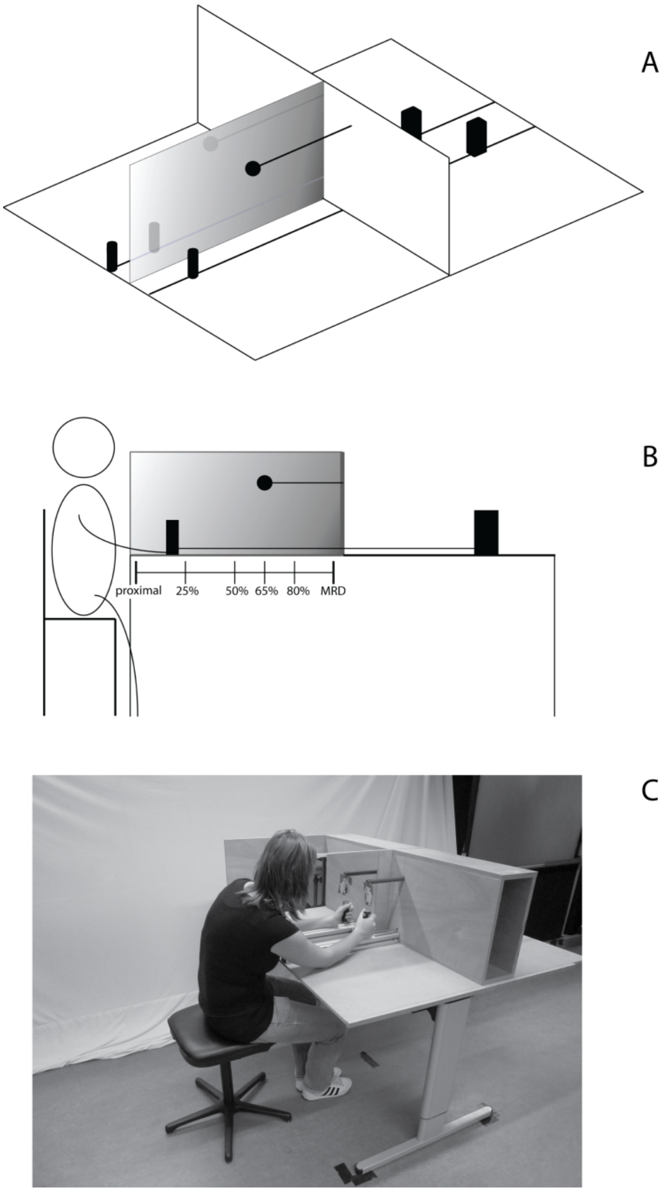

Figure 5.1: (A) Top view of the experimental setup with the two handles that could be moved back and forth along the track. The divide between the arms was either an opaque screen or a mirror. The position of the handles outside the box was measured with an Optotrak camera (not depicted here). (B) Side view of the experimental setup. The proximal starting position and the four target positions (25%MRD, 50%MRD, 65%MRD, 80%MRD) are indicated. Note that the target positions were determined based on the maximum reaching distance of each child and thus differed per participant. (C) Real-life picture of the experimental setup.

Results

All 23 participants were able to complete the experiment according to the instructions and all participants could perform a bimanual symmetrical movement as indicated by the small differences in starting time between the arms (difference between arms in mirror condition: M = -0.05 sec, SD = 0.25, t_{22} = -0.96, p = 0.035; difference between arms in screen condition: M = 0.04 sec, SD = 0.28, t_{22} = 0.74, p = 0.47). Although slightly larger, the differences in end time between the arms were also relatively small (difference between arms in mirror condition: M = 0.48 sec, SD = 1.19, t_{22} = 1.95, p = 0.06; difference between arms in screen condition: M = 0.59 sec, SD = 0.94, t_{22} = 3.04, p = 0.006).

Nevertheless, one trial was excluded because participant 15 did not perform a symmetrical bimanual movement, i.e. the movement of the impaired hand was initiated after the movement of the less-impaired arm was finished. In addition, 14 out of 368 trials in the bimanual condition had to be excluded from the analysis (PP 1 [2 trials], 3 [4], 15 [3], 8 [2], 23 [2], 12 [1]) because the less-impaired arm was not on the target location at the end of the movement. In case the difference between less-impaired arm and target was more than half of the distance between two consecutive target locations, the trials could not be assigned to either target distance and therefore they were excluded from analysis. This exclusion of trials meant that for some participants the value for a certain condition was based on one trial instead of the mean of two trials.

Unimanual vs. bimanual task (impaired arm)

Matching accuracy

Matching accuracy differed significantly between the unimanual and the bimanual task, and a significant Task by Distance effect indicated that this difference was distance dependent ($F_{3,66}$ = 3.16, p = 0.03; see Figure 5.2). Absolute error was smaller in the bimanual task compared with the unimanual task for all but the 25%MRD target position. In addition, absolute error was found to increase with increasing distance for both the unimanual and the bimanual task. However, between 50% and 65% and between 65% and 80%MRD the increase in error was not significant for either task.

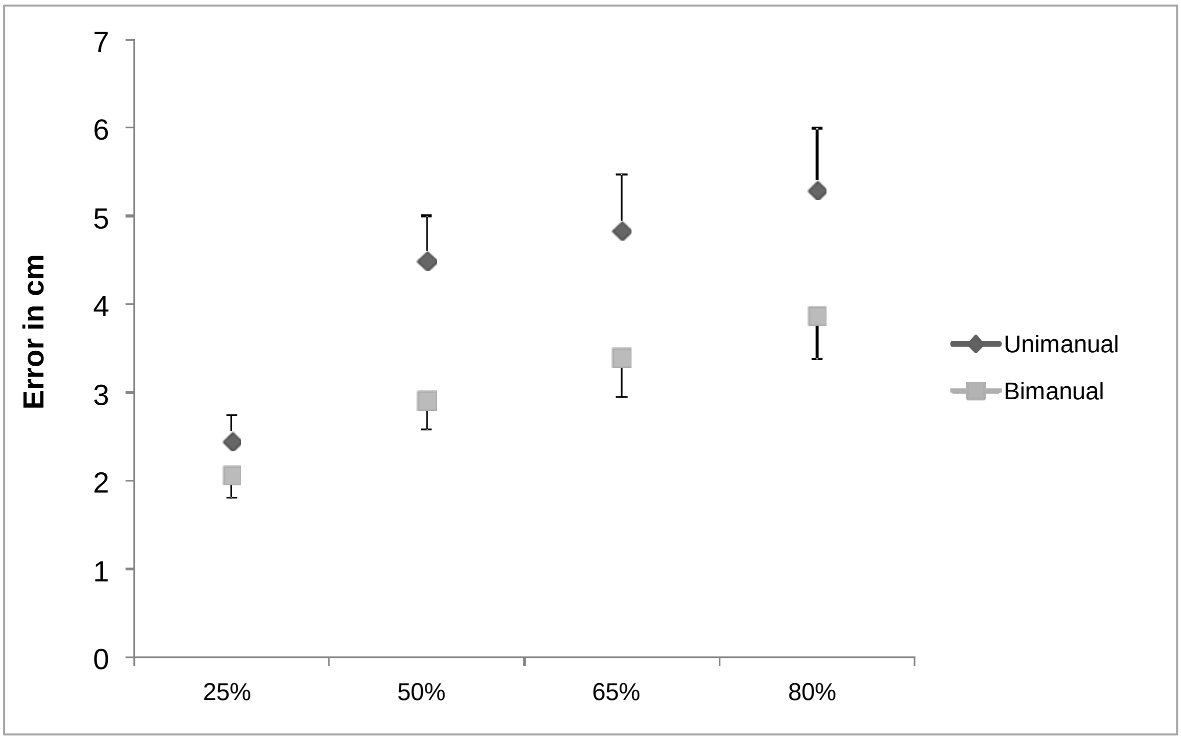

Figure 5.2: The absolute error (in cm; mean and SE values) increased with increasing distance (25%, 50%, 65%, 80%MRD on the horizontal axis) for both the unimanual (dark grey) and the bimanual task (light grey).

Despite the significant Task-effect (unimanual vs. bimanual) on matching accuracy at group level, close inspection of the individual data showed that the advantage of moving simultaneously with the two hands was not present in all participants. In 14 out of 23 individuals absolute error in the bimanual condition was smaller than in the unimanual condition for 3 or 4 of the 4 target distances (see Table 5.2; *Bi+ group*). However, for both the *Bi+* and the *Bi- group* it was demonstrated that the absolute error in the unimanual condition was positively correlated with the size of the decrease in error in the bimanual condition (Table 5.3), i.e. a larger error in the unimanual condition was related to a greater improvement in the bimanual condition.

Furthermore, as the repeated measures ANOVA showed, there was no effect of Visual condition on matching accuracy of the bimanual task (i.e. no interaction effect between Visual condition and Task), thus mirror visual feedback of the target did not affect absolute error. Inspection of the individual data of the bimanual task, however, indicated that in 13 out of 23 participants absolute error was smaller in the mirror condition compared to the screen for 3 or 4 of the 4 distances (see Table 5.2; *Mirror+ group*). In Figure 5.3 the mean errors in the screen and the mirror condition are depicted for the *Mirror+* and the *Mirror-* group.

In order to reveal whether this variability in response to mirror visual feedback was related to the proprioceptive accuracy of the impaired arm when no mirror visual feedback was available, we examined for both groups (*Mirror*+ and *Mirror*-) the correlation between the error in the screen condition ('baseline condition') and the improvement in accuracy due to the mirror, i.e. the difference in error between the screen and the mirror condition. Table 5.4 shows these correlations and the corresponding *p*-values for the *Mirror*+ and the *Mirror*- group. No significant correlations were found for the *Mirror*- group, whereas significant positive correlations between the baseline error and the improvement in accuracy due to the mirror were observed for the *Mirror*+ group on all four distances. This suggests that for individuals who do better in the mirror than in the screen condition in the majority of the target distances (*Mirror*+ *group*), a larger error in the screen condition is related to a larger decrease in error in the mirror condition, i.e. to a higher degree of improvement in the mirror condition.

In addition, we examined with a Mann-Whitney U test whether the *Mirror*+ *and Mirror*- group differed in terms of scores on the MACS, WeeFIM and Tardieu scale. No differences between the groups were found for the MACS (z = -0.69, p = 0.52; mean rank *Mirror*+ = 12.81, *Mirror*- = 10.95) and the WeeFIM (z = -0.40, p = 0.74; mean rank *Mirror*+ = 11.54, *Mirror*- = 12.60). However, the *Mirror*+ group showed a higher average Tardieu score when compared to the *Mirror*- group (1.65 and 1.17 respectively; z = -2.17, p = 0.04; mean rank *Mirror*+ = 13.88, *Mirror*- = 8.06). However, no significant correlation was found between the degree of improvement (mean improvement over the four distances) in the mirror condition and the Tardieu score for the *Mirror*+ group (Spearman's rho = -0.34, p = 0.26), the *Mirror*- group (Spearman's rho = 0.36, p = 0.34) and both groups together (Spearman's rho = 0.32, p = 0.15).

*Table 5.2: Classification of the participants into groups. For each participant and each target distance (25%, 50%, 65%, 80%MRD) an asterisk (*) indicates when the error was smaller in the bimanual condition compared to the unimanual condition (left part of the table) and when the error was smaller in the mirror compared to the screen condition in the bimanual condition only (right part of the table). When in 3 or 4 out of 4 distances the error was smaller in the bimanual condition, the participant was assigned as performing better in the bimanual condition compared to the unimanual condition (Bi+). For the screen/mirror comparison the same principle was used. When the error was smaller in the mirror condition compared to the screen condition (indicated with *) the participant was assigned to the Mirror+ group (i.e. Mirror+ = +).*

P	***Bi*+ vs. *Bi*-**					***Mirror*+ vs. *Mirror*-**				
	25%	**50%**	**65%**	**80%**	***Bi*+ or *Bi*-?**	**25%**	**50%**	**65%**	**80%**	***Mirror*+ or *Mirror*-?**
1	*	*		*	+	*	*	*	*	+
2	*			*	-	*	*	*	*	+
3		*	*	*	+			*	*	-
4	*	*	*		+	*	*		*	+
5		*		*	-	*		*	*	+
6	*	*	*	*	+	*				-
7	*	*	*	*	+	*		*	*	+
8	*	*	*	*	+	*	*	*	*	+
9					-	*				-
10	*	*	*	*	+	*				-
11			*	*	-		*			-
12		*	*	*	+					-
13	*	*	*	*	+	*		*		-
14		*			-		*	*	*	+
15		*		*	-		*	*	*	+
16	*	*	*	*	+	*	*	*	*	+
17	*	*	*	*	+	*		*	*	+
18	*	*	*	*	+	*	*	*	*	+
19	*	*	*	*	+		*	*	*	+
20	*	*	*	*	+	*	*		*	+
21		*			-			*		-
22			*	*	-		*			-
23		*	*		-			*		-

Average velocity

There was no effect of Task on average velocity ($F_{1,21} = 0.45$, $p = 0.51$; Unimanual = 5.1 cm/s, Bimanual = 4.8 cm/s). Moreover, Visual condition did not have an effect on the average velocity ($F_{1,21} = 1.25$, $p = 0.28$; Mirror: 4.7 cm/s Screen: 5.2 cm/s). However, a significant main effect of Distance was found ($F_{1.76,\, 38.61} = 30.40$, $p < 0.001$), indicating an increase in velocity with increasing distance (25%: 3.50 ± 0.32 cm/s; 50%: 4.83 ± 0.47 cm/s; 65%: 5.45 ± 0.59 cm/s; 80%: 5.99 ± 0.52 cm/s).

Movement smoothness

A main effect of Distance ($F_{2.33,\, 51.26} = 57.03$, $p < 0.001$) and a significant interaction effect between Task and Distance were found ($F_{1.92,\, 42.27} = 60.21$, $p = 0.005$) for movement smoothness. No differences between the unimanual and the bimanual task were found on all of the four distances. However, for both the unimanual and the bimanual task the relative number of velocity peaks decreased (i.e. movement smoothness increased) with increasing distance (except for the unimanual task between 50% and 65%MRD and for the bimanual task between 65% and 80%MRD).

Table 5.3: For each distance the correlations are reported between the error in the unimanual task (U25, U50, U65, U80) and the difference in error between the unimanual and the bimanual condition, i.e. error in the unimanual condition minus the error in the bimanual condition (DifUB25, DifUB50, DifUB65, DifUB80) for the Bi+ and the Bi- group. The table shows Pearson's r value and the corresponding p-value. Significant correlations are indicated with an asterisk.

Group	**Correlation**	**Pearson *r***	***p*-value**
Bi+ (n=14)	U25 vs. DifUB25	0.51	0.16
	U50 vs. DifUB50	0.98	<0.001*
	U65 vs. DifUB65	0.73	0.03*
	U80 vs. DifUB80	0.61	0.08
Bi- (n=9)	U25 vs. DifUB25	0.36	0.21
	U50 vs. DifUB50	0.74	0.002*
	U65 vs. DifUB65	0.72	0.003*
	U80 vs. DifUB80	0.76	0.002*

Table 5.4: For each distance the correlations are reported between the error in the screen condition (S25, S50, S65, S80) and the difference in error between the screen and the mirror condition, i.e. error in screen condition minus the error in mirror condition (DifMS25, DifMS50, DifMS65, DifMS80) for the Mirror+ and the Mirror- group. The table shows the Pearson's r value and the corresponding p-value.

Group	**Correlation**	**Pearson *r***	***p*-value**
Mirror+ (n=13)	S25 vs. DifMS25	0.69	0.009*
	S50 vs. DifMS50	0.76	0.002*
	S65 vs. DifMS65	0.70	0.007*
	S80 vs. DifMS80	0.69	0.009*
Mirror- (n=10)	S25 vs. DifMS25	0.13	0.73
	S50 vs. DifMS50	-0.007	0.99
	S65 vs. DifMS65	0.31	0.39
	S80 vs. DifMS80	-0.43	0.22

Bimanual task

In order to examine differences in kinematics between the impaired and the less-impaired arm, a repeated measures ANOVA was performed with Visual condition (mirror, screen), Distance (25%, 50%, 65%, 80%MRD) and Arm (impaired, less-impaired) as within factors. Moreover, in order to examine differences between the *Mirror+* and the *Mirror-* group this factor (Mirror-group) was included as between factor in the 3-way repeated measures ANOVA.

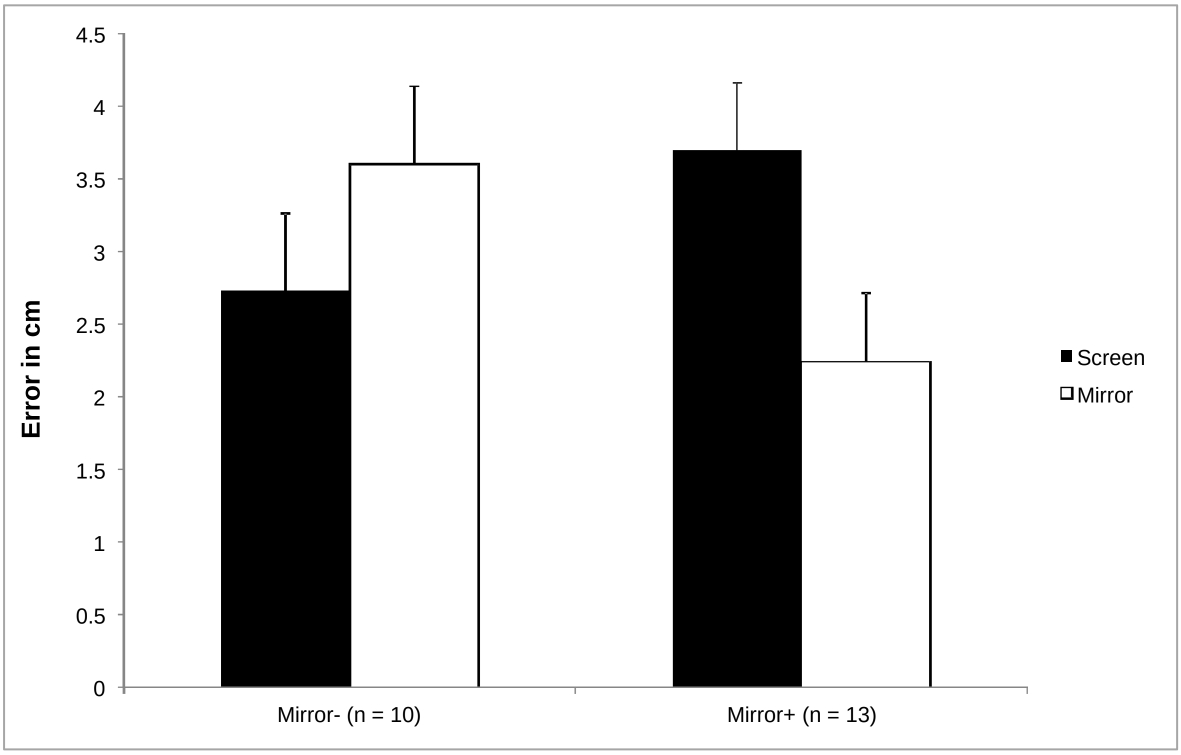

Figure 5.3: Absolute error (in cm) for the Mirror+ and the Mirror- group in the screen and the mirror condition. The Wilcoxon signed rank test revealed a significantly higher error in the screen compared to the mirror condition for the Mirror+ group (z = -3.18, p < 0.00). For the Mirror- group the error was higher in the screen compared to the mirror condition (z = -2.50, p = 0.01).

Average velocity

The ANOVA revealed a significant main effect of Visual condition ($F_{1,21} = 5.84$, $p = 0.03$). The average velocity was 0.7 cm/sec lower in the mirror condition (4.6 ± 0.5 cm/s) compared to the screen condition (5.3 ± 0.7 cm/s). Furthermore, the significant main effects of Arm ($F_{1,21} = 5.14$, $p = 0.03$) and Distance ($F_{1.95,\,41.03} = 21.22$, $p < 0.001$) were modified by a significant interaction effect between Arm and Distance ($F_{2.57,\,53.90} = 9.62$, $p < 0.001$) and a significant interaction between Arm, Distance, and Mirror-group ($F_{3,63} = 3.16$, $p = 0.03$; see Table 5.5).

Inspection of the 3-way interaction showed no differences between the *Mirror+* and the *Mirror-* group. For both the *Mirror+* group and the *Mirror-* group and both arms a significant increase in $V_{average}$ was found when the distance that had to be covered increased. Moreover, comparing the average velocity between the impaired and the less-impaired arm showed for both groups higher velocities in the less-impaired than in the impaired arm, but only for larger distances (65% and 80%MRD).

Movement smoothness

The analysis of the relative movement smoothness revealed a significant effect of Distance ($F_{1.86,\ 38.97} = 33.96$, $p < 0.001$) and a significant interaction effect between Arm and Distance ($F_{1.71,\ 36.00} = 3.76$, $p = 0.04$). For both the impaired and the less-impaired arm the relative number of velocity peaks decreased (i.e. movement smoothness increased) with increasing distance (except for the 65% to 80%MRD). In addition, the number of velocity peaks was higher in the impaired compared to the less-impaired arm, indicating a lower relative movement smoothness for the impaired arm, but only for the 80%MRD (impaired arm = 0.32 peaks/cm vs. less-impaired arm = 0.25 peaks/cm).

Table 5.5: Mean and SE values for the $V_{average}$ and Movement Smoothness. Values are given for each distance (25%, 50%, 65%, 80%MRD) in the unimanual and bimanual movement condition for the impaired and the less-impaired arm. Note that no values are reported for the less-impaired arm in the unimanual condition because this task was not performed in the present study.

		Unimanual	**Bimanual**	
	Distance	**Impaired arm**	**Impaired arm**	**Less-impaired arm**
$V_{average}$ (cm/s)	25%	3.71 ± 0.40	3.19 ± 0.40	3.16 ± 0.34
	50%	4.88 ± 0.52	4.79 ± 0.56	5.20 ± 0.59
	65%	5.54 ± 0.57	5.36 ± 0.73	6.08 ± 0.82
	80%	6.19 ± 0.56	5.80 ± 0.63	6.40 ± 0.64
Movement smoothness (peaks/cm)	25%	0.68 ± 0.069	0.51 ± 0.074	0.57 ± 0.088
	50%	0.43 ± 0.048	0.39 ± 0.071	0.38 ± 0.073
	65%	0.37 ± 0.044	0.33 ± 0.049	0.28 ± 0.054
	80%	0.30 ± 0.042	0.32 ± 0.071	0.25 ± 0.051

Discussion

This study examined the difference in matching accuracy of the impaired hand between a unimanual and a bimanual condition and the effects of mirror visual feedback on matching accuracy in children and adolescents with SHCP. Consistent with earlier studies that showed beneficial effects on the timing and the control of the impaired hand and arm when moving the two hands simultaneously (e.g. Steenbergen et al., 1996; Sugden & Utley, 1995; Utley & Sugden, 1998), we found a significant increase in matching accuracy (37.5% on average) in the bimanual condition compared to the unimanual condition. In addition, mirror visual feedback led to better matching in 13 out of 23 participants. Together, these findings support the application of bimanual symmetrical movements and the use of mirror visual feedback in the treatment of upper limb function, though additional research is warranted to determine under what circumstances and for whom this approach is effective.

The underlying mechanism of the improved matching accuracy in the bimanual condition is probably related to facilitative processes resulting from bilateral connections throughout the central nervous system. For example, neural crosstalk is suggested to constrain homologous muscle groups to act as a single coordinative structure during bimanual symmetrical movements, which enhances the coupling between the limbs and also more abstract parameters (e.g. amplitude, force, direction; Cattaert, Semjen, & Summers, 1999; Swinnen & Wenderoth, 2004). In addition, we suggest that in the present study congruent visual and proprioceptive information of the less-impaired arm, which was available in the bimanual condition and presumably served as a frame of reference, may have facilitated accurate placement of the impaired arm (see also Smorenburg et al., 2011).

Consistent with other research (Ledebt et al., in preparation; Smorenburg et al., 2011, 2012; van Beers, Sittig, & Denier van der Gon, 1998), larger errors were made in (unimanual and bimanual) matching movements with larger amplitude. Note that larger movements were also relatively faster and smoother. This counterintuitive finding for this population suffering from spasticity may be explained by the rather slow overall speed of movement execution. Spastic movement disruptions are commonly observed at higher speeds, and in this self-paced task it is likely that participants avoided detrimental effects of spasticity.

Concentrating on the effects of mirror visual feedback, the results of the present study showed that both hands moved slower in the mirror condition compared to the screen condition. Further, there was no improvement in accuracy of the impaired hand when mirror visual feedback of the less-impaired hand was available. Remarkably though, inspection of individual data revealed a positive effect of mirror visual feedback on matching accuracy in a considerable number of individuals (13 out of 23). In fact, mirror visual feedback seemed to hamper accurate placement of the impaired arm in the remainder of the group, which may explain the absence of a statistical effect at group level.

Explaining the mechanisms underlying the positive effect of the mirror remains speculative, but using transcranial magnetic stimulation (TMS) and advanced brain imaging techniques in healthy individuals, researchers have begun to uncover the neural basis of the mirror effects. For example, Garry, Loftus, and Summers (2005) have shown that the excitability of the ipsilateral[1] primary motor cortex (M1) is facilitated when healthy adults were viewing a mirror reflection of the moving hand (see also Nojima et al., 2012; Tominaga et al., 2011). In addition, mirror visual feedback was found to alter touch perception by enhancing the tactile sensitivity in the ipsilateral posterior parietal cortex (PPC; Ro, Wallace, Hagedorn, Farne, & Pienkos, 2004) and, further, to lead to increased activation within the ipsilateral superior temporal gyrus (STG; Matthys et al., 2009).

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[1] Ipsilateral refers to the hemisphere at the same side of the moving arm which was visible in the mirror.

Finally, the findings of Hamzei et al. (2012) suggest a remodelling of the motor system with a pivotal role for the contralateral[2] sensorimotor cortex (SMC) after training with the mirror (see also Michielsen et al., 2011). Apparently, mirror visual feedback has the capacity to induce plastic changes in brain regions directly involved in motor control (M1, SMC) and regions that have been linked with the mirror neuron system (PPC, STG).

The involvement of (part of) these specific regions might also (partly) explain the variability in response to mirror visual feedback across individuals. Staudt et al. (2002) found that the SHCP population may be functionally classified on the basis of the size of the lesion. Larger lesions are accompanied with a cortical reorganisation of the primary motor cortex and premotor areas towards the contralesional cortex, whereas no reorganisation is observed when the lesion is small. Wilke et al. (2009) on the other hand, found that the primary sensory cortex was preserved in the contralateral, lesioned hemisphere, irrespective of the extent of the lesion, which means that the sensorimotor control loop is disrupted when motor areas are relocated to the contralesional side. This variety in clinical picture might then be related to the variability in behavioural response to mirror visual feedback found in the current study. The idea that heterogeneity in patient groups, and more in particular variance in the neural resources, can explain the varying success of interventions is consistent with earlier findings in individuals with SHCP or a hemiparesis after stroke (McCombe Waller & Whitall, 2008; Ramachandran & Altschuler, 2009).

Our findings highlight that it is essential to determine which children might benefit most from therapy with mirror visual feedback e.g. by using data on the side of the lesion or corticospinal reorganisation. Unfortunately, lack of brain imaging and other neurophysiological data do not allow us to identify in which particular groups of children and adolescents mirror visual feedback may be favourable. However, behavioural evidence indicates that the extent of improvement in the mirror condition is related to the size of the error in baseline conditions. A similar result was found for the improvement under bimanual conditions, i.e. the improvement was larger when the error in the unimanual condition was greater. Both bimanual practice and practice with the mirror thus seem to be more effective in individuals with more severe problems of position sense. Still, it is possible that the children, who did not show an improvement in the mirror condition at present, need more practice before effects can be detected. A higher level of spasticity also seemed to be related to the efficacy of the mirror, given that the *Mirror+* group showed higher levels of spasticity of the *Mirror-* group. However, the difference between the two

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[2] Contralateral refers to the hemisphere contralateral to the moving arm which was visible in the mirror.

groups was very small and no significant correlations were found between the degree of improvement and the Tardieu score. Moreover, it is questionable whether a (coarse) clinical measure for spasticity can be related to a sensitive measure for position sense as used in the present study.

In conclusion, the current study showed that for children and adolescents with hemiplegia matching with the impaired hand is more accurate in a bimanual than in a unimanual matching condition. Similarly, mirror visual feedback had a positive effect on movement accuracy of the impaired arm, however, only in a subset of the individuals with SHCP. This variability in response may be related to differences in size and location of the brain lesions of the CP population and/or to the initial position sense of the impaired arm. Further research examining the relation between spasticity, position sense and improvements due to mirror visual feedback together with advanced brain imaging is warranted to determine which children might benefit most from bimanual practice with mirror visual feedback.

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Appendix

The Tardieu Scale measures spasticity using two parameters: the spasticity angle and the spasticity grade (Gracies et al., 2010). The spasticity angle is the difference between the angles of arrest at slow speed and of catch-and-release at fast speed. The spasticity grade is an ordinal variable that grades the intensity and measures the muscle’s reaction to fast passive stretch.

In this study we used the spasticity grade as an indication for the level of spasticity. Gracies et al. (2010) showed for this measure high intrarater and interrater reliability for experienced raters; 90% ± 8% and 81% ± 13% respectively.

The Functional Independence Measure for children (WeeFIM) includes 18 items covering six areas in two dimensions (i.e. motor and cognitive). Motor: self-care (eating, grooming, bathing, dressing upper body, dressing lower body, toileting); sphincter control (bladder management, bowel management); transfer (chair/bed/wheelchair transfer, toilet transfer, tub/shower transfer); locomotion (crawling/walking/wheelchair, stair climbing). Cognitive: communication (comprehension, expression) and social cognition (social interaction, problem solving, memory; Sperle, Ottenbacher, Braun, Lane, & Nochajski, 1996; Tur et al., 2009). In the present study we only used the motor items of the WeeFIM. Ottenbacher et al. (1996) showed high test-retest responses for the WeeFIM with an intraclass correlation coefficient of 0.97.

The Manual Ability Classification System (MACS) is designed to classify how children with CP use their hands for object handling in daily life (Eliasson et al., 2006). It reports the collaboration of both hands together and is not an assessment of each hand separately. As shown in the study of Eliasson et al. (2006), the MACS has a good validity and reliability: intra-class correlation coefficient between therapists was 0.97.

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Chapter 6

Practicing a matching movement with a mirror in individuals with spastic hemiplegia: from visual to proprioceptive control of movement?

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Abstract

Individuals with spastic hemiparetic cerebral palsy (SHCP) have proprioceptive deficits, which hamper them to perform and to learn new tasks. Mirror visual feedback has been shown to improve movement performance in individuals with SHCP. Therefore, the current study examined the effect of practice of a matching task with (mirror) visual feedback of the less-impaired arm on the matching accuracy of the impaired arm in this patient group.

The practice consisted of 40 trials of bimanual target matching, where one group received regular visual feedback and a second group received mirror visual feedback of the less-impaired arm. On three occasions (pre, post, and after a one-week-retention) position sense of the impaired arm was tested with a unimanual and bimanual matching task, performed without any visual information of either hand. Matching accuracy of the impaired arm was higher in the post-test than in the pre-test, but this improvement was similar for both training groups. In the retention-test, accuracy had returned to pre-test-level, which might be ascribed to the short duration of the training. These outcomes suggest that practicing a matching task with visual feedback of the less-impaired arm might help to improve the matching accuracy of the impaired arm in SHCP.

Introduction

Proprioception can broadly be described as the sense of body parts in space, which is an important aspect in the control of movement. Proprioception consists of two components: position sense (sense of static position) and kinesthesia (sense of movement). In children and adolescents with Spastic Hemiparetic Cerebral Palsy (SHCP) both components of proprioception are deteriorated compared to typically developing peers (Chrysagis, Skordilis, Koutsouki, & Evans, 2007; Goble, Hurvitz, & Brown, 2009; Smorenburg, Ledebt, Deconinck, & Savelsbergh, 2012a; Wann, 1991; Wingert, Burton, Sinclair, Brunstrom, & Damiano, 2009). Individuals with this congenital disorder show spasticity and motor impairments lateralized to one side of the body as a result of a unilateral lesion in the developing foetal or infant brain (Krägeloh-Mann & Staudt, 2008). To the best of our knowledge, it has not been examined whether proprioception in children with SHCP, and more specifically the position sense of the impaired arm, is susceptible to practice.

Research has shown that during motor development and learning, a shift in reliance from visual to proprioceptive control takes place (Fleishman & Rich, 1963; Smyth & Marriott, 1982). The visual control of the effector is important early in learning whereas the monitoring of the limbs is delegated to proprioception as learning proceeds. In children with SHCP this shift from visual to proprioceptive control is expected to be hampered considerably due to disturbed proprioception and increased reliance on visual feedback (Verrel, Bekkering, & Steenbergen, 2008). Therefore, any therapeutic intervention that aims to improve motor function with the involvement of visual feedback in children with SHCP depends on its effect on proprioception.

Recently, mirror visual feedback (i.e. mirror therapy) has been introduced as a possible way to improve motor function of individuals with SHCP (Feltham, Ledebt, Bennett, Deconinck, Verheul, & Savelsbergh, 2010; Feltham, Ledebt, Deconinck, & Savelsbergh, 2010; Gygax, Schneider, & Newman, 2011). However, the effects of mirror visual feedback on position sense in this population remain unknown. Mirror visual feedback is generated by placing a mirror in between the arms in the sagittal plane. When the participant looks into the mirror from the less-impaired side, the mirror image of the less-impaired arm is superimposed on the impaired arm and the illusion is created that both arms are moving in perfect symmetry. Smorenburg, Ledebt, Deconinck and Savelsbergh (2012b) suggested that movement accuracy[1] of the impaired arm may be improved by

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[1] Proprioception is a difficult concept to measure. Therefore, researchers often fall back on the assessment of position sense which can be measured with a position matching task. It is generally well accepted that the magnitude of the matching error, i.e. matching accuracy can be a useful indicator of the proprioceptive acuity and is thus used as outcome variable. (Goble, 2010)

moving bimanually with mirror visual feedback of the less-impaired arm. In their study, participants moved towards a target either with the impaired arm only (unimanual) or with both arms symmetrically (bimanual). Vision of the impaired arm was blocked by an opaque screen in between the arms, but the less-impaired arm was always visible. Smorenburg et al. 2012b demonstrated that the matching error of the impaired arm decreased when moving in symmetry with the less-impaired arm, compared to when moving only with the (invisible) impaired arm. Moreover, for a subset of the participants with SHCP, mirror visual feedback of the less-impaired arm improved the movement accuracy of the impaired arm during the bimanual condition compared to ‘regular’ visual feedback of the less-impaired arm (screen condition). Consequently, with the present study we aimed to examine whether the proprioceptive component of a movement can be practiced in individuals with SHCP by repetitively performing a matching movement with mirror visual feedback of the less-impaired arm.

Methods

Participants

The participants for this study were recruited in 2 schools for special education in The Netherlands (Werkenrode school, Groesbeek and De Piramide, The Hague). From the seventeen children with SHCP that participated in the study, 16 children were included for analysis (15.8 ± 2.5 years; 3 females; see Table 6.1). One participant dropped out after less than half of the training because he was too fatigued. The participants did not have a visual impairment (which was not corrected to normal), pain in either of the upper limbs, Botox treatment in the past six months preceding the measurement or any other neuromuscular disorder than CP. All participants understood the basic instructions in order to perform the measurement. An indication of the severity of the children’s impairment is provided by means of the Tardieu score for spasticity (Gracies et al., 2010), the Functional Independence Measure for children (WeeFIM; Sperle, Ottenbacher, Braun, Lane, & Nochajski, 1996), and the Manual Ability Classification System (Eliasson et al., 2006; MACS; Table 6.1). Participant’s parents provided written informed consent prior to testing. All procedures were approved by the institutional research ethics committee and in accordance with the Declaration of Helsinki.

Table 6.1: Participant characteristics. For each participant the age (in years), sex (Male, Female), Impaired arm, Tardeu score, WeeFIM and MACS score, aethiology and Maximum reaching distance (MRD) of the impaired and less-impaired arm (in cm) are given. The last column (C) represents the number of completed practice trials and the condition in which the trials were practiced (i.e. Mirror-group = M; Screen-group = S).

P	Age (years)	Sex	Impaired arm	TS[a]	WeeFIM/MACS	Aetiology	MRD I/LI	C
1	14.3	M	Left	0.5	91/1	Unknown	30.5/32	M40
2	15.3	M	Left	1	91/2	Cerebral infarction	33/36	S35
3	17.6	M	Left	2	91/1	Premature	37/38	M40
4	17.7	M	Left	1.5	91/2	Unknown	40/46	M35
5	13.7	M	Left	1	91/1	Perinatal origin	40/40	S40
6	19.3	M	Left	1	88/2	Right cerebral infarction	39/41	M35
7	18.3	M	Left	1	90/1	Perinatal cerebral infarction	37/41	S30
8	15.1	M	Left	1.5	59/3	Streptococcen infection at 5 weeks	37/41	S30
9	13.2	F	Left	1	89/2	Unkown	25/28	S20
10	16.4	M	Left	1	62/3	Schizencephaly right	39/46.5	S30
11	15.2	M	Right	1.5	91/2	Premature	26/28	M40
12	13.0	F	Right	1	90/2	Unknown	23/28	S30
13	18.0	M	Right	1.5	91/2	Unkown	24/42	S20
14	16.8	F	Right	0.5	91/1	Perinatal asphyxia	29/30	M35
15	19.3	M	Right	1	91/3	Premature (twins)	30/34	S40
16	10.3	M	Right	1	91/1	Unkown	29/37	M30

[a]TS = Tardieu score for spasticity; mean of the individual scores for the Biceps and the Triceps.

Procedure of pre-test, post-test and retention-test

Matching accuracy was measured pre, post and after one-week retention. The post-test was performed immediately after the training, after a 5-10 minute break. The retention-test was performed exactly one week after the post-test.

In order to do so, children were seated on a height adjustable chair behind a height adjustable table with the knees flexed to 90°. On the table a custom made wooden box was placed with two handles in a slit, one at each side of an opaque divide, running parallel in the sagittal and horizontal plane (Figure 6.1). The handles were located 20 cm apart and the maximum anterior-posterior range was 56 cm. The handles inside the box were attached to two handles outside the box on which light emitting diodes were attached. One unit with three infrared cameras (3020 Optotrak Northern Digital Inc., Waterloo, Canada) was used to measure the position of the markers at a sample rate of 200 Hz. An opaque sheet was placed on top of the arms (not touching the arms) so that they were not visible during the movement. Before the start of the measurement, the maximum reaching distance was determined (MRD). For this, the participant was asked to grasp the handles and extend the elbows as far a possible without bending forward. The MRD of the impaired arm was used to calculate the different target positions to be used in the test and practice. If a participant was unable to grip the handle due to physical impairment, the experimenter placed the hand on top of the handle. The test consisted of a unimanual and a bimanual matching task, the order of which was randomly assigned to the participants.

In the unimanual task the participants were asked to move the handle towards a target with the impaired or with the less-impaired arm. Target positions were scaled to the individual's MRD and were located at 20%, 40%, 60%, 70%, and 80%MRD. With each arm, two trials per target position[2] (i.e. 10 trials per arm in total) were performed. The trials were grouped into two blocks, one for each arm, with the target positions randomised within one block. The procedure of the bimanual task was the same, except for the fact that participants were instructed to move the two handles to the target with the impaired and the less-impaired arm simultaneously and in a symmetrical fashion. Two trials per target position were executed.

Procedure of the practice period

The practice of the matching task was performed after the pre-test, varying from one day to one week. In this training the participants were instructed to perform bimanual symmetrical matching movements towards a target placed at 40% or 60%MRD. The hand started either from a proximal (with the handle at 0%MRD) or distal position (at 100%MRD). The different combinations of target position (2) and starting position (2) were randomly presented to the participants and repeated ten times resulting in total number of 40 trials. A short break was given after 20 trials. The participants were randomly allocated to one of the two training groups. One training group (mirror group; n = 7) practiced the bimanual movements with mirror visual feedback, i.e. a mirror was placed in between the arms and so that the participant saw the less-impaired arm and its mirror reflection. The other group (screen group; n = 9) practiced the movement with an opaque screen in between the arms, so that visual feedback of the less-impaired arm only was available. For both groups the impaired arm was invisible. After each practice trial the experimenter provided feedback (knowledge of results; KR) indicating the size of the endpoint error made by the impaired arm (see below), both verbally (e.g. 'you are 3 cm from the target') and visually by scaling her fingers. In addition, proprioceptive feedback was given by passively moving the impaired arm to the target location so that the participant could 'feel' the correct location. Since not all children were able to complete

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[2] The post-test was an exception to this. There, each target was presented only once. This was decided based on the fatigue of the participants and time constraints.

the total of 40 training trials due to fatigue or concentration problems, Table 6.1 reports the number of training trials completed by each participant. For the purpose of analysis, the training was divided into three parts, irrespective of the total number of trials that was executed. The first part of the training consisted of the first 5 trials, the middle part of the training consisted of the middle 5 trials of the training and the last part of the training consisted of the last 5 trials of the training.

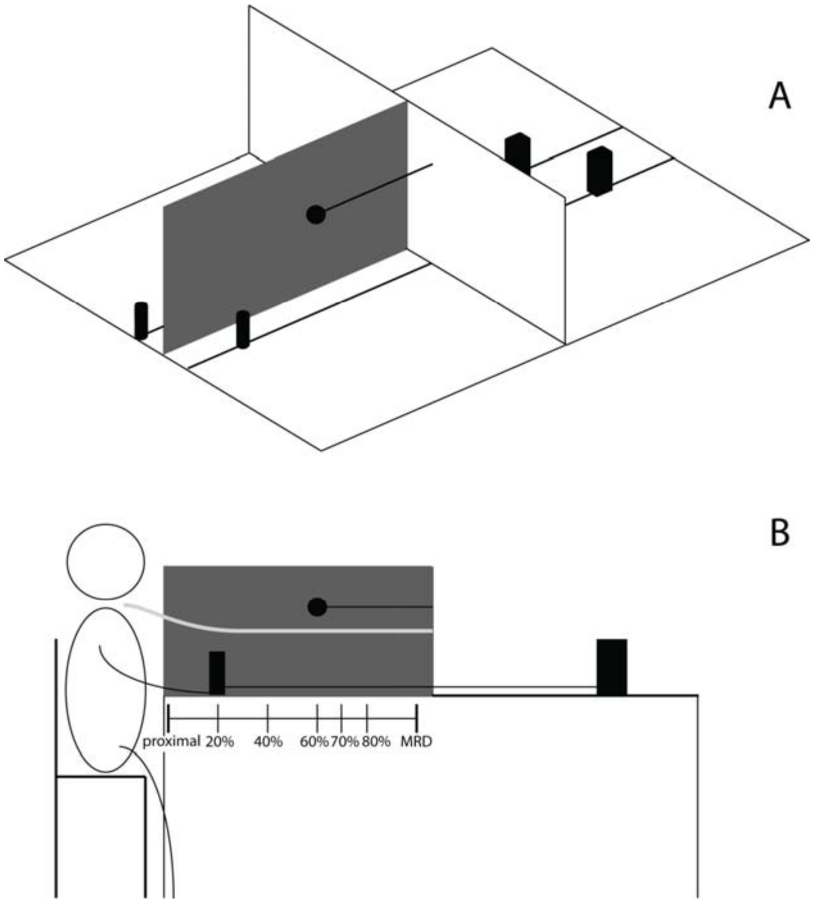

Figure 6.1: Experimental setup. (A) Top view of the setup. (B) Side view of the setup with the different target positions (20%, 40%, 60%, 70%, 80%MRD). The hands of the participants were covered by an opaque sheet in the pre-, post-, and retention-test (grey line). The target is depicted as a circle.

Data analysis

Pre- and post-training and after a retention period of 1 week, absolute error of the impaired and the less-impaired arm was calculated using custom-written Matlab routines (The Mathworks, version 2011) for both the unimanual tasks and the bimanual task. The absolute error corresponds to the distance in cm between the target and the position of the (less-) impaired arm at the end of the movement. The end of the movement was determined as the moment where the movement velocity dropped below 5 mm/s (van Roon, Steenbergen, & Meulenbroek, 2005).

Statistical analysis

Two sets of analysis were conducted. The aim of the first analysis was to determine whether the period of practice was effective in improving overall position sense and to check if mirror visual feedback resulted in larger gains. We therefore created an overall error score, i.e. the mean absolute error of the impaired arm averaged across the 5 target positions and across the two tasks (unimanual and bimanual matching). Then a repeated measures ANOVA was conducted on the overall error score with Test moment (pre-test, practice phase (early, mid, late), post-test and, retention-test) and Arm (impaired vs. less-impaired arm) as a within factor and Training group (mirror vs. screen) as a between factor. Secondly, a repeated measures ANOVA was conducted to study the effect of Arm (impaired vs. less-impaired), Task (unimanual vs. bimanual), Target location (20%, 40%, 60%, 70%, and 80%MRD) and Training group (mirror vs. screen) on the matching accuracy in the pre- and the post-test. The significance level was set at 0.05. In case sphericity assumptions were violated, Greenhouse-Geisser adjustments were applied. Fisher’s LSD test was used for post-hoc comparisons.

Results

Effects of practice

A significant effect of Test moment indicates a positive influence of the practice period on the matching accuracy ($F_{2.36,\,33.0} = 14.01$, $p<0.001$). For both training groups a significantly larger error in the pre-test (no visual information) compared to the post-test was found, suggesting that matching accuracy of the impaired arm improved after a period of practice. After the retention period, however, mean absolute error returned to the level of the pre-test.

Mean absolute error during the practice period was smaller than for all three tests (pre-, post-, and retention-test), indicating that adding visual information of the less-impaired arm and KR had an immediate positive effect. During the training the error of both groups decreased significantly (from 1.71 cm in the first part to 1.23 cm in the last part; p = 0.001). Finally, the analysis revealed a main effect of Arm ($F_{1,14}$ = 13.53, p = 0.002), showing that overall the impaired arm (2.89 ± 0.31 cm) had larger errors than the less-impaired arm (1.82 ± 0.11 cm).

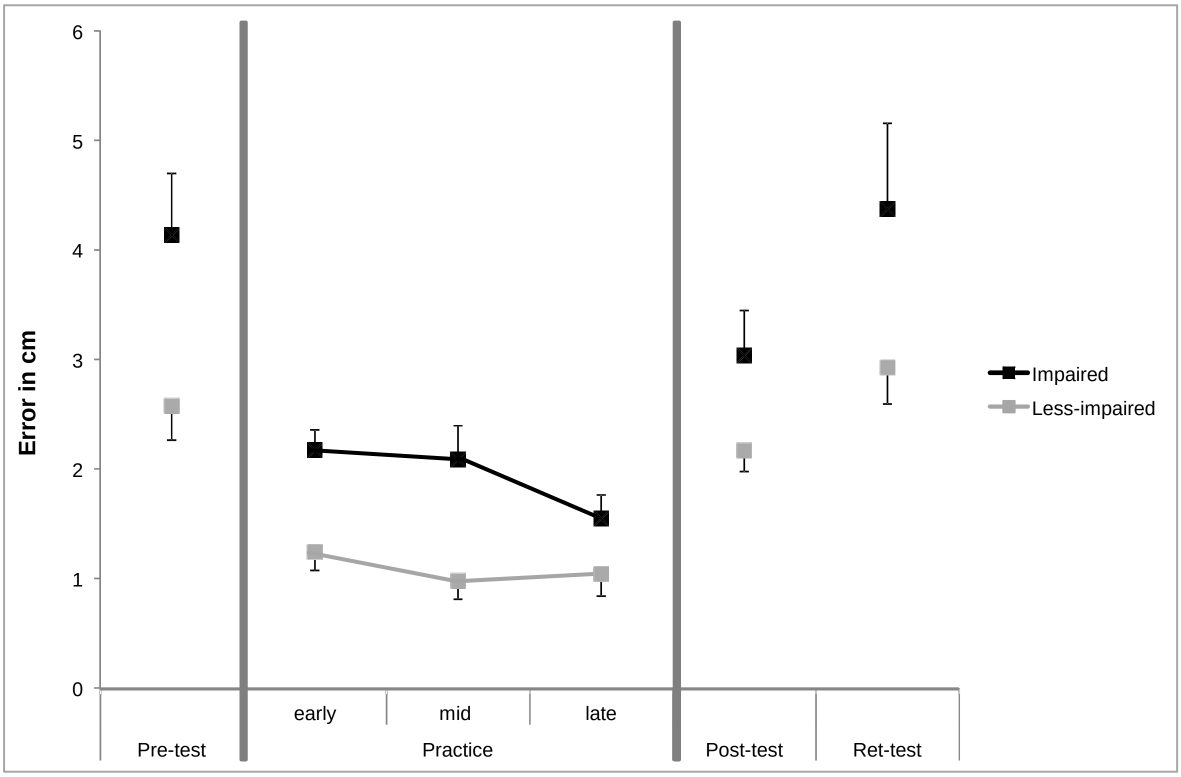

Figure 6.2: Absolute error for the impaired and the less-impaired arm in the pre-test, training parts (early, middle, late), post-test and retention-test.

Effects of Arm, Task, Distance and Training group on the accuracy in pre- and post-test

This analysis revealed a main effect of Arm ($F_{1,14}$ = 12.08, p = 0.004) and a main effect of Test ($F_{1,14}$ = 7.65, p = 0.015), which were combined into a significant Arm x Test interaction effect ($F_{1,14}$ = 4.88, p = 0.044). Post-hoc analysis demonstrated for the impaired arm a significant decrease in error between the pre-test and the post-test, whereas the error of the less-impaired arm was the same on both test moments (see Figure 6.2). Moreover, it was found that the error in the impaired arm was always greater than the error in the less-impaired arm. No effects of Target location were observed (i.e. no differences in absolute error between the 5 target locations). Finally, a main effect of Task was found ($F_{1,14}$ = 5.27, p = 0.038), demonstrating that the error in the unimanual task (2.79 ± 0.30 cm) was significantly smaller than the error in the bimanual task (3.17 ± 0.35 cm).

Discussion

The present study examined the effects of practicing a matching task on the matching accuracy of the impaired arm in children and adolescents with SHCP and looked for potential differences between practice with and without mirror visual feedback of the less-impaired arm. A positive effect of the practice on position sense was found. After practice, endpoint error of the impaired arm had dropped with 26.6% but the error increased again to the level at pre-test after a 1 week retention period. Moreover, it is interesting to note that this training effect not just occurred for the target positions that were practiced (i.e. 40% and 60%MRD). This implies that the effect of practice is not distance specific and suggests a transfer so that position sense is improved over a larger range of motion.

Although the overall effect of the training was positive, no differences were found between the screen-practice group and the mirror-practice group. It thus seems that mirror visual feedback of the less-impaired arm (i.e. ‘illusory’ visual feedback of the impaired arm) does not provide extra information to improve matching accuracy of the impaired arm as compared to ‘regular’ visual feedback of the less-impaired arm. This seems to be in contrast with previous findings showing positive effects of mirror visual feedback on movement accuracy in SHCP (Smorenburg et al., 2012b), although it must be noted that Smorenburg et al. did show that only a subset of the individuals with hemiplegia benefited from the mirror. Post-hoc inspection of the current individual data indicated a decrease in the overall error score after practice (pre vs. post-test) in 5 out 7 participants of the mirror-group and 8 out of 9 participants of the screen-group. We thus see some variation in response to the training with and without mirror visual feedback, but overall there is a positive effect of practice. As suggested by Smorenburg et al. (2012b) the variation might be due to the nature and the severity of the brain lesion, but attention might also be a confounding factor as suggested by Moseley et al. For some participants looking towards the impaired arm (i.e. seeing the mirror reflection of the less-impaired arm) might augment attention towards the impaired arm, which in turn enhances the learning process (Moseley & Wiech, 2009). For others, focusing attention on the mirror reflection might have perturbed sensory-motor integration due to problems with dividing attention over multiple processes or a decreased sense of agency of the movement seen in the mirror (Moseley & Wiech, 2009). However, the lack of a difference between the two practice groups might also be due to the nature of the feedback during the practice period. Participants received verbal/visual feedback about the size of the error and the impaired arm was passively displaced to the correct position. This combination of feedback might have turned away the learning effects from the mirror and might be another reason for not finding a difference between the two methods of practice.

The decrease in error of the impaired arm after practice with (mirror) visual feedback of the less-impaired arm and KR suggests that a transfer from visual to proprioceptive control occurred during learning. Although this is in line with earlier studies on motor learning (Adams, Gopher, & Lintern, 1977; Fleishman & Rich, 1963), this study is, to the best of our knowledge, the first to show that this transfer can even occur for individuals with deficits in position sense (Chrysagis et al., 2007; Goble et al., 2009; Smorenburg et al., 2012a; Wingert et al., 2009) and a high dependence on visual information (Verrel et al., 2008). This has important implications for therapy. Bimanual movement coordination has been shown to be deteriorated in individuals with SHCP when compared to typically developing individuals (Hung, Charles, & Gordon, 2004). If practice can lead to a more proprioceptive control of movements and less visual control is needed, this might facilitate the bimanual coordination so that activities of daily living can be performed more effectively. We cannot ascribe the improved accuracy in the present study to the availability of visual feedback only, since the participants also gained KR. However, given the immediate decrease in error in the early practice phase with visual feedback of the less-impaired arm (compared to the pre-test without visual information) it can be suggested that the congruent visual and proprioceptive information of the less-impaired arm served as a frame of reference. The participant learned to link the visual and proprioceptive information which in turn helped to improve (the use of) the position sense of the impaired arm and decreased the reliance on visual information as learning proceeds. However, the positive effect of practice was not present on the retention-test. Therefore, longer training experiments should verify whether indeed a (long-term) transfer in movement control takes place in this patient group and whether this can lead to improvements in everyday functioning.

In conclusion, the current study showed that practice of a matching movement with visual feedback of the less-impaired arm together with KR temporarily improved position sense of the impaired arm in children with spastic hemiplegia. At this moment the effects of practice cannot be ascribed to mechanisms that are particularly related to mirror visual feedback, but it seems that active practice of a matching movement with visual feedback can reduce the dependence on visual feedback in individuals with SHCP.

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Chapter 7

General discussion

Finishing this thesis I realised that after three years of PhD-research I have more questions than when I started. Nevertheless, the work in this thesis provided more insight into the previously reported positive effects of mirror visual feedback in children with SHCP and the visuo-proprioceptive interactions in children and adolescents with SHCP. In this final chapter of my thesis I will first briefly explain the main findings of each chapter. Subsequently, I will discuss the results and elaborate further on the implications of our findings. Finally, I will provide ideas for future studies based on the work in this thesis.

Main findings

The study described in chapter two elaborated upon the experiments of Feltham, Ledebt, Bennett et al. (2010) and Feltham, Ledebt, Deconinck et al. (2010). From their studies it was unclear whether the positive effects of mirror visual feedback on neuromuscular activity and bimanual symmetry were the result of viewing a symmetrical movement (irrespective of which arm was viewed) or were the result of the illusion that the impaired arm had been substituted by the less-impaired arm. Therefore we investigated in **chapter two** the effect of (mirror) visual feedback of the *impaired arm* on the neuromuscular activity and the movement symmetry. It was found that the amount of neuromuscular activity in the Biceps muscle of the impaired arm was higher when receiving mirror visual feedback of the impaired arm than receiving mirror visual feedback of the less-impaired arm. No effects on movement kinematics were found. This suggests that the effects reported by Feltham, Ledebt, Bennett et al. (2010) and Feltham, Ledebt, Deconinck et al. (2010) are likely not caused by the perception of two symmetrically moving limbs per se, but by the illusion that the impaired arm is substituted.

Chapter three aimed to get more insight into deficits in position sense in children with SHCP when compared to typically developing (TD) children for a task that involve both arms. To this end, a contralateral matching task was performed. We found that children with SHCP have difficulties matching the position of one arm with the position of a static reference arm (without any visual information available) when compared to TD children. Moreover, the matching accuracy was lower when the distance that had to be covered by the matching arm was larger.

In **chapter four** we examined the effect of (mirror) visual feedback of the non-moving (reference) arm on the matching accuracy of the moving arm in children with SHCP. When participants looked into the mirror they saw their static arm and its mirror reflection, which created the illusion that both arms were already at the target position. It was demonstrated that static (mirror) visual feedback improved the matching accuracy of the moving arm compared to a situation without visual information. Moreover, a similar distance effect was found as in chapter three: a larger distance to target resulted in a lower matching accuracy.

In **chapter five** we examined the effect of moving the impaired arm in synchrony with the less-impaired arm. In addition, the effect of mirror visual feedback on the matching accuracy of the impaired arm was investigated. We showed that the accuracy of the impaired arm improved when moving in synchrony with the less-impaired arm, than when moving alone. Furthermore, we demonstrated that mirror visual feedback in the bimanual movement condition can lead to a greater matching accuracy of the impaired arm for a subset of the individuals with SHCP. For this group, a poorer position sense in a condition without mirror visual feedback was related to greater improvements in accuracy when mirror visual feedback was available.

Finally, **chapter six** was designed to examine the effects of practicing a matching movement with (mirror) visual feedback of the less-impaired arm on the matching accuracy of the impaired arm in individuals with SHCP. Overall, a positive effect of the practice with visual feedback was found. That is to say, the matching error was smaller in the post-test when compared to the pre-test. However, practice with the mirror did not seem to have a differential effect on the accuracy than training with ‘regular’ feedback of the less-impaired arm. Nevertheless, practicing a matching movement with visual feedback seems to induce a transfer from visual to proprioceptive control of movement.

Position sense in individuals with SHCP

Given the important role of proprioception in motor control, the effectiveness of any therapeutic intervention that aims to improve motor function in SHCP is partly dependent on its effect on proprioception. Mirror visual feedback might be a possible tool for rehabilitation and therefore the work in this thesis examined the effects of mirror visual feedback on the static component of proprioception, position sense. A number of studies already showed an impaired position sense in SHCP by actively moving one limb towards a visible or remembered target (Chrysagis, Skordilis, Koutsouki, & Evans, 2007; Goble, Hurvitz, & Brown, 2009; Wingert, Burton, Sinclair, Brunstrom, & Damiano, 2009). However, the ability to match the position of one arm by actively moving the other arm had not been considered. With a contralateral matching task we demonstrated that individuals with SHCP are clearly disadvantaged for the accurate positioning of one arm relative to the position of the other arm, which is required in multiple manual tasks.

But what causes the impaired proprioception in individuals with SHCP? Is there a deficit on the peripheral level (i.e. sensory system and/or muscle) so that perturbed signals are sent from the muscle to the brain? And/or, are the signals unperturbed and lies the problem in the processing of these signals in the brain (i.e. a problem on central level)? Malformation and injury to cortical and subcortical structures as the parietal lobe and the thalamus are believed to impair sensation (Clayton, Fleming, & Copley, 2003 In: Majnemer, Bourbonnais, & Frak, 2008). However, in chapter three we stated that if the matching difficulties can be explained by a deficit on cortical level only, this would result in distance independent matching errors for both arms. We found that matching error increased when the distance to cover was larger, so it seems that in children with SHCP deficits occur not only at central level but more likely at both the central and the peripheral level.

Focusing on the peripheral level, spasticity is a major symptom of SHCP which may affect position sense. Spasticity causes the muscle to be shortened and stiffened (Friden & Lieber, 2003), which may increase or disturb the discharge of the muscle spindles (Wingert et al., 2009). This would suggest that higher levels of spasticity would result in larger proprioceptive impairments. However, in chapter four we did not find a clear relationship between spasticity and matching accuracy and also in chapter five the relation between spasticity and improvement in accuracy due to the mirror was inconclusive. Chrysagis and colleagues (2007) on the other hand reported a significant negative relation between the degree of spasticity (measured with the Modified Ashworth Scale) and the position sense. Differences in velocity between the matching movements and the measurement of the spasticity might be a confounding factor in this respect. Moreover, it is questionable whether (coarse) clinical scales for spasticity can be related to sensitive measures of position sense. Thus, although the exact role of the spastic muscle in the proprioceptive deficits remains to be determined, it is conceivable that deficits on muscle level also contribute to the position sense deficits in certain circumstances.

Some caution is warranted when interpreting the results of position matching experiments. There are several ways to measure position sense, but the matching error might be influenced by different factors (Goble, 2010). One of these factors (and maybe the most important one) is the type of matching task. The choice for a particular matching task might seem trivial but the characteristics of the task can greatly influence your results. In the ipsilateral paradigm, the participant needs to memorize the target position before matching it with the same (ipsilateral) hand. It is likely that in these situations, part of the matching error measured is due to cognitive or memory deficits rather than a decrease in position sense. The contralateral (concurrent) matching task used in the present thesis (chapters three and four) eliminates the involvement of memory, but has limitations of its own. Because of the involvement of both arms, it is difficult to ascertain from which arm the matching error arises (reference arm, matching arm or both). Moreover, matching with the opposite limb requires greater inter-hemispheric transfer, as proprioceptive information from one limb likely crosses the hemispheric divide through the transcallosal pathways of the corpus callosum. This could lead to increased cognitive load that might influence the matching error (Goble, 2010). For healthy children, adolescents and elderly, no significant differences between these two matching tasks were found (Adamo, Martin, & Brown, 2007; Goble, Lewis, Hurvitz, & Brown, 2005; Ledebt, Smorenburg, & Savelsbergh, submitted), but for individuals with asymmetric brain injuries and memory problems the matching errors can be greatly influenced by the type of task used.

A transfer from visual to proprioceptive control of movement?

In healthy individuals it has been suggested that during learning a shift from visual to proprioceptive control takes place. In the early stages of learning visual control is dominant whereas in later stages of learning, people rely more on proprioceptive information (Fleishman & Rich, 1963; Smyth & Marriott, 1982). Moreover, it has been suggested that higher sensitivity to proprioceptive cues could facilitate this transfer of control (Fleishman & Rich, 1963). In individuals with SHCP this transfer is expected to be considerably hampered due to an increased reliance on visual information and a disturbed proprioception, which can have a detrimental effect of the efficacy of e.g. mirror therapy. Therefore, we examined in chapter six whether in individuals with SHCP the control of movement can be delegated from vision to proprioception during practice of a matching movement. If matching accuracy improves (i.e. smaller errors on the proprioceptive post-test) after a period of practice with visual information, this could suggest that the sense of limb position is modulated which in turn facilitates the proprioceptive control of movement. Indeed, we demonstrated that learning a matching movement with both arms in synchrony under visual control of the less-impaired arm, led to smaller matching errors of the impaired arm when the movement was subsequently performed without visual information (proprioceptive control only). This is in agreement with studies on motor learning in TD-individuals (Adams, Gopher, & Lintern, 1977; Fleishman & Rich, 1963). However, to the best of our knowledge this is the first study that showed that this transfer can also occur in individuals with impaired proprioception and increased reliance on visual information.

How can we explain the change in movement accuracy we observed? First of all it has been suggested that during learning vision improves accuracy by providing a detailed spatial structure (i.e. frame of reference) for the storage of movement-related information (Laabs & Simmons, 1981 In: Proteau, Marteniuk, Girouard, & Dugas, 1987). Indeed, as shown in chapter four and six, vision seems to play an important role given the fact that adding visual information led to a greater accuracy than matching with proprioceptive information only. Although we cannot ascribe the improvement in accuracy to visual feedback of the less-impaired arm only (we also provided knowledge of results), the immediate decrease in error from pre-test to the first phase of practice suggests that visual feedback plays an important role in the improvement after practice and possibly in the transfer to a more proprioceptive control of movement. In addition, as Wong, Wilson and Gribble (2011) suggested, improvements in proprioception after motor learning could reflect a sensory component of short-term sensorimotor plasticity that occurred during learning (see also: Ostry, Darainy, Mattar, Wong, & Gribble, 2010). Motor learning is dependent upon plasticity in the motor areas, but changes in proprioception have been found in conjunction with improvements in motor performance. It is therefore suggested that motor learning can modify both the motor areas and the somatosensory systems (processing of somatosensory information), which is visible in increased matching accuracy after practice. This possible link between motor learning and sensory changes could lead to novel approaches to rehabilitation for individuals with SHCP (Wong et al., 2011).

The effects of mirror visual feedback in SCHP

The studies presented in this thesis used mirror visual feedback for two purposes: on the one hand we used the mirror as a tool to manipulate visual feedback of the position of the matching hand to examine visuo-proprioceptive interactions in a contralateral matching task (i.e. both hands seemed to be on the endpoint position already at the beginning of the movement). On the other hand we explored the possibilities to use the mirror for therapy purposes in individuals with SHCP, as described in chapter five and six. In chapter four, we examined whether the illusion that both hands were already at the endpoint position could alter the perceived location of the hand behind the mirror. We showed that mirror visual feedback of a static reference arm did not alter the matching accuracy when compared to the screen condition in which only the reference arm was visible. In other words, the participants were, despite the illusion, able to sense the position of their hidden limb. In contrast with the results of Holmes and Spence (2005), the illusion created by mirror visual feedback in the present study did thus not influence the matching accuracy of the participants. However, in the study of Holmes and Spence (2005) the position of the reference hand was manipulated in the medio-lateral plane whereas in the present study the bias was created in the anterior-posterior plane. Furthermore, Holmes and Spence (2005) showed that a longer exposure time to the mirror increasingly biased the endpoint error towards the direction specified by the mirror visual feedback. The short exposure time to the mirror before the start of the movement might be the reason for the fact that the conflict situation in the present study did not affect the matching accuracy.

The positive findings in a range of patients with acquired unilateral motor and/or pain disorders suggests that mirror therapy may be a suitable method to improve upper limb function. However, it is still unclear whether the positive effects of mirror therapy in patients with acquired disorders can be extrapolated to individuals with unilateral congenital disorders such as SHCP. The work presented in this thesis followed on the work of Feltham, Ledebt, Bennett et al. (2010) and Feltham, Ledebt, Deconinck et al. (2010) who showed for a bimanual symmetric inward circular movement that the presence of mirror visual feedback led to decreased interlimb movement variability (i.e. more stable pattern of movement symmetry) and decreased levels of eccentric neuromuscular activity in the Biceps muscle of the impaired arm. These effects were immediate, i.e. the children were exposed to mirror visual feedback for 2 minutes and within this time frame the effects were visible. More recently, Gygax and colleagues (2011) examined the effects of a period of training with mirror visual feedback in children with SHCP. After a 3-week training consisting of three repetitive symmetrical upper limb exercises either with or without mirror visual feedback (divided over two groups; cross-over design), improvements were reported in grasp strength and the position of the upper limbs during achievement of specific tasks (dynamic position analysis measured with the SHUEE evaluation). The work in this thesis added to the existing body of knowledge by showing that mirror visual feedback can enhance matching accuracy in individuals with SHCP, which is an indicator of position sense. However, in chapter five we showed that mirror visual feedback seems to improve matching accuracy of the impaired arm for a subset of the participants only. This variability in responsiveness to the mirror is interesting because until now, no studies examined/mentioned the possibility that mirror visual feedback might only be suitable for a part of the CP-population. Correspondingly, Ramachandran and Altschuler (2009) pointed out in their review article that the variability in results in stroke patients suggests that the procedure of mirror visual feedback might help some patients more than others. They proposed as well that this variability in stroke patients may depend in part on the exact location of the lesion. Our study together with the notion of Ramachandran and Altschuler thus indicates that more research is warranted in order to establish which individuals will benefit from therapy with mirror visual feedback. On the other hand, one could argue that there is no reason why mirror therapy should not be implemented routinely given the simplicity of the procedure (Ramachandran & Altschuler, 2009). Still, in daily practice it takes a lot of time before the best suitable therapy for a patient is found; time that could have been spend to actually improve arm/hand functionality. It would therefore be very useful to know which therapy might be most suitable for a specific individual.

In line with our suggestions, Kuhnke et al. (2008) showed differential efficacy of a 12 day constraint-induced movement therapy (CIMT) protocol for different types of corticospinal reorganisation in SHCP (identified by transcranial magnetic stimulation). They showed a lower efficacy of CIMT for patients whose paretic hand is controlled by the ipsilateral (i.e. contralesional) hemisphere (ipsi-group) than for patients whose paretic hand is controlled by the contralateral (i.e. lesioned) hemisphere (contra-group). Two possible reasons for the differential effect of CIMT have been put forward by the authors. First, it is suggested that CIMT 'rebalances' the (disbalanced) interhemispheric inhibition in hemiparesis (i.e. in SHCP the more active contralesional hemisphere inhibits the activity in the less active affected hemisphere). Constraining the less-affected arm can reduce the cortical activity in the contralesional hemisphere and intensive repetitive training of the impaired arm can increase the cortical activity in the affected hemisphere. However, since in the ipsi-group the motor representations of both the impaired and the less-impaired arm are located in the same hemisphere, targeting interhemispheric inhibition with CIMT is thought to be ineffective. Bimanual therapy might be a better option for this group. Second, in the ipsi-group the sensorimotor loop is disrupted, as S1 is located in the lesioned hemisphere, whereas for the contra-group this sensorimotor loop is preserved (with M1 receiving immediate somatosensory feedback from the moving hand via S1; see also Wilke et al., 2009). This intact sensorimotor loop might be crucial for effective motor learning during CIMT and can thus explain the differences in efficacy of CIMT between the groups. Although this study focused only on the effects of CIMT, the results of this study confirm the suggestions that there is an interaction between treatment type and corticospinal reorganisation in SHCP. Further research is thus warranted to investigate this for other treatment types, such as mirror therapy.

Underlying mechanisms of mirror visual feedback

As suggested in chapter five mirror visual feedback might be a possible tool for rehabilitation for a *subset* of the individuals with SHCP rather than for the population as a whole. Moreover, in the previous paragraph some neurological evidence is provided for dissociation in therapy effects within one patient group. Nonetheless, before being able to draw conclusions on which patients will benefit from mirror therapy and why, it is also necessary to get more insight into the working mechanisms of mirror visual feedback. Although unravelling the underlying mechanisms of mirror visual feedback was not within the scope of my thesis, I would like to discuss the different hypotheses that have been put forward in the literature (see Ramachandran & Altschuler, 2009 for a review).

First of all, it has been suggested that the mirror might restore the congruence between discrepant visual feedback and motor output leading to an unlearning of (learned) non-use in unilateral disorders like stroke. Ramachandran (2005) assumes that, at least part of, the (learned) paretic movement in stroke can be attributed to a discrepancy between the internal copy of the motor command sent by the central nervous system (i.e. efference copy) and the afferent sensory information. When the motor commands are not confirmed by the proprioceptive feedback, motor output is amplified which is believed to further deteriorate motor performance. Mirror visual feedback may help to restore the congruence between the two systems.

Another hypothesis focuses on the mirror neurons, a network of neurons in the parietal and frontal lobe of the brain, which is activated when observing or imaging motor tasks and is involved in action planning (Rizzolatti & Craighero, 2004). It is suggested that mirror visual feedback might activate (dormant) mirror neurons in the damaged parts of the brain thereby facilitating neural plasticity or revival. This in turn could improve movement on the impaired side of the body.

A third mechanism that has been put forward is the (enhanced) recruitment of ipsilateral pathways. Most of our motor function is controlled by corticospinal tracts that are crossed over at the level of the medulla oblongata and therefore the right hemisphere controls the left side of the body and the left hemisphere controls the right side of the body. However, a small portion of the tract does not cross over. These tracts are called the ipsilateral pathways. In healthy individuals the majority of these ipsilateral pathways is withdrawn during the perinatal period, but when there is a damage which impairs the contralateral tract to function properly, e.g. as a result of cerebral damage, the ipsilateral pathways may persist. Staudt and colleagues (2002) showed differences in the amount of ipsilateral projections in SHCP depending on the size of the lesion: individuals with large lesions did not have any contralateral projections but instead only showed ipsilateral projections. Individuals with small lesions only showed preserved contralateral projections and no functional ipsilateral projections. Mirror visual feedback is suggested to act upon the ipsilateral pathways but it remains to be determined if this actually happens and in what way.

Finally, Garry, Loftus and Summers (2005) showed that the excitability of the primary motor cortex ipsilateral to the moving hand was facilitated significantly more in the condition with mirror visual feedback of the moving hand than in the other conditions. According to the authors, this increased M1 excitability could lead to practice-induced neuroplasticity within the affected M1 in patients with a unilateral brain lesion (Garry et al., 2005; Ramachandran & Altschuler, 2009). In summary, the range of positive findings in patients with (acquired) unilateral motor problems (Feltham, Ledebt, Bennett et al., 2010; Feltham, Ledebt, Deconinck et al., 2010; Gygax et al., 2011; McCabe et al., 2003; Sathian, Greenspan, & Wolf, 2000) suggest that mirror therapy may be a suitable method for improvement of upper limb function. Still, the underlying mechanisms of mirror therapy remain poorly understood.

Future directions

The experiments in this thesis were designed to get more insight into the proprioceptive abilities and the visuo-proprioceptive interactions (i.e. the effects of (mirror) visual feedback on the proprioceptive abilities) in individuals with SHCP. This research, however, was a first step and future research is warranted to unravel the sensory problems of individuals with spastic hemiplegia and to determine how we can use mirror visual feedback in the therapy regime of this patient population.

Mirror therapy

First of all, research should elaborate further on the effects of mirror visual feedback (mirror therapy) on motor performance in individuals with spastic hemiplegia. Different studies showed positive effects of mirror therapy in different unilateral patient groups and, although limited, the first results in children with SHCP are promising. Moreover, mirror therapy is easy to apply, inexpensive and non invasive and as such may be considered an interesting complement to the rehabilitation of children and adolescents with spastic hemiplegia (not excluding other established forms of therapy; Gygax et al., 2011). I therefore believe that the use of mirror visual feedback in therapy for SHCP deserves further attention. Future research is needed to confirm the positive effects of this treatment on different areas of motor control and sensation (Gygax et al., 2011). Furthermore, as mentioned earlier in this thesis (chapter five), it is suggested that not all children will benefit (to the same extent) from mirror therapy due to e.g. differences in size and location of the brain lesion. It is therefore recommended to determine which children might benefit from practice with mirror visual feedback and for what reason. Extensive documentation on the characteristics of the disorder in each individual might help in this respect. Studies incorporating brain imaging techniques such as TMS or (f)MRI might provide us with more insight into different types of brain reorganisation and the relation to the efficacy of a therapy. Moreover, it can be interesting to see whether cortical reorganisation occurs after prolonged training with mirror visual feedback.

On the level of therapy implementation there are also certain aspects that deserve attention. For example, it remains to be determined whether mirror therapy can function as a therapy on its own, or whether some children would benefit more by first ‘jump-starting’ with e.g. CIMT (Gordon & Steenbergen, 2008) followed by mirror therapy, or the other way around. In order to start with CIMT a certain level of functionality is needed, but for mirror therapy no such requirements are set (yet). It is possible that mirror therapy is effective to get some movement in a spastic arm which can then be followed by another period of therapy with e.g. CIMT or HABIT. In this respect, it is interesting to note that little is known about the effect of mirror training in severely affected individuals since most studies to date have focused on mildly to moderately impaired individuals. In order to know for which patients mirror therapy might be most effective it is therefore unavoidable to examine the effects in severely afflicted individuals as well. Moreover, it is necessary to determine the timing and modalities of intervention (Gygax et al., 2011). What kind of tasks should be incorporated in the training, what should be the intensity of the training and what are the long-term effects (determined in a large patient population with a Randomised Controlled Trial)? Eventually detailed guidelines could be developed for accurate application of mirror therapy in SHCP.

Accuracy vs. precision

A limitation of the studies in this thesis was that we only focused on the absolute error. Although it is generally accepted that absolute matching error is a useful indicator for deficits in position sense, the precision of a matching task might also provide insight into the noise within the information processing system, which can arise from the sensory signals or from the processing of these signals (van Beers, Sittig, & Denier van der Gon, 1998).

Active vs. passive examination

The majority of the studies that examined position sense in TD individuals or patients with SHCP used active matching tasks, requiring the generation of a motor command. It is worth noting that Paillard and Brouchon (1968) showed smaller errors in an active matching task than in a passive matching task in a small group of healthy adults. Apparently position sense is more accurate under active than under passive conditions in healthy individuals and signals related to motor commands also contribute to position sense (see also Gandevia, Smith, Crawford, Proske, & Taylor, 2006). It is unknown, however, if this is also the case in individuals with a unilateral brain damage and muscle spasticity. It is therefore recommended to investigate the contribution of 'passive' receptors and 'active' motor commands to position sense in individuals with SHCP. A similar contralateral matching task as used in chapters three and four could be used. In the active condition the participant actively moves one arm until both arms are at the same position. In the passive condition the arm is moved passively towards the target position and the participant has to indicate when both arms are at the same position.

Proprioception vs. somatosensation

In this thesis the focus is on one aspect of proprioception, the position sense. However, I would like to stress that proprioception is not the same as somatosensation. The term somatosensation encompasses both the proprioceptive sensation (i.e. kinaesthesia and position sense) and the cutaneous sensation (e.g. tactile discrimination, vibration perception and texture discrimination). Somatosensory impairments may modulate motor performance, and it is therefore essential to evaluate these as part of the rehabilitation management of children with neurological conditions such as CP. Rather than focusing only on position sense, it might thus be interesting to examine the cutaneous sensation in conjunction with the proprioceptive sensation. Previous research showed for individuals with SHCP impairments on the level of stereognosis and two-point discrimination in conjunction with deficits in pressure sensitivity, vibration sense and directionality (see Majnemer et al., 2008 for a review). Although there is a paucity of studies that actually looked at the relationship between sensation and hand function in SHCP, it can be assumed that impaired cutaneous sensation also affects motor performance in children with SHCP (Auld, Boyd, Moseley, Ware, & Johnston, 2012a; Tachdjian & Minear, 1958).

In order to be able to programme planning and selection of therapeutic approaches to optimize function it is crucial to get a comprehensive documentation of the extent and the range of somatosensory impairments in these children. However, due to a lack of (reliable) tools, and the fact that feasibility of accurate assessment of sensory abilities is constrained by physical, cognitive and behavioural impairments (assessment of many modalities requires good attention and concentration skills), it is challenging to assess the sensory impairments in children and youth with SHCP (Majnemer et al., 2008). Despite these demerits, it is recommended to focus future research on the assessment of the different components of proprioception and cutaneous sensation in this patient group and examine the effects on and the relationship with arm/hand functionality. For example, a weight differentiation experiment, in which the detection threshold is determined, can give more insight into the 'overall' proprioceptive ability in individuals with SHCP. A more detailed insight into the different abilities can be obtained by specific tests for each sensory modality (e.g. two-point discrimination, stereognosis, position sense, kinaesthesia, pressure sensitivity). Understanding the nature and the severity of the impairments in proprioception and cutaneous sensation in individuals with SHCP might assist to direct treatment to improve sensation but might also facilitate the overall rehabilitation process in terms of learning new motor tasks (Auld, Boyd, Moseley, Ware, & Johnston, 2012b).

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Appendix A

List of publications

Papers included in the thesis

Smorenburg, A.R.P., Ledebt, A., Deconinck, F.J.A., & Savelsbergh, G.J.P. (submitted). Practicing a matching movement with a mirror in individuals with spastic hemiplegia: from visual to proprioceptive control of movement?

Smorenburg, A.R.P., Ledebt, A., Deconinck, F.J.A., & Savelsbergh, G.J.P. (2012). Matching accuracy in hemiparetic cerebral palsy during unimanual and bimanual movements with (mirror) visual feedback. *Research in Developmental Disabilities, 33(6),* 2088-2098.

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Smorenburg, A.R.P., Ledebt, A., Feltham, M.G., Deconinck, F.J.A., & Savelsbergh, G.J.P. (2011). The positive effect of mirror visual feedback on arm control in children with Spastic Hemiparetic Cerebral Palsy is dependent on which arm is viewed. *Experimental Brain Research, 213 (4),* 393-402.

Smorenburg, A.R.P., Ledebt, A., Deconinck, F.J.A., & Savelsbergh, G.J.P. (2011). Visual feedback of the non-moving limb improves active joint-position sense of the impaired limb in children with Spastic Hemiparetic Cerebral Palsy. *Research in Developmental Disabilities, 32 (3),* 1107-1116.

Other publications by the author

Deconinck, F.J.A., Benham, A., Feltham, M.G., **Smorenburg, A.R.P.,** Ledebt, A., & Savelsbergh, G.J.P. (in preparation). Is mirror therapy, the new holy grail for hemiplegic cerebral palsy? A critical review of current evidence.

Ledebt, A., **Smorenburg, A.R.P.,** & Savelsbergh, G.J.P. (in preparation). Younger children show less accurate and distance-dependent upper limb matching ability compared to adolescents.

Ledebt, A., **Smorenburg, A.R.P.,** & Savelsbergh, G.J.P. (in preparation). Age- related differences in upper limb matching accuracy in children and young adults are task dependent.

Van Kampen, P.M., Ledebt, A., **Smorenburg, A.R.P.,** Vermeulen, R.J., Kelder, M.E., van der Kamp, J., & Savelsbergh, G.J.P. (2011). Gaze behaviour during interception in children with Spastic Unilateral Cerebral Palsy. *Research in Developmental Disabilities, 33 (1), 45-53.*

Conference proceedings

Smorenburg, A.R.P., Ledebt, A., Deconinck, F.J.A., Savelsbergh, G.J.P. Accuracy of unimanual vs. bimanual matching in individuals with Spastic Hemiparetic Cerebral Palsy. *8th FENS forum of neuroscience.* Barcelona, Spain, 14-18 July 2012.

Smorenburg, A.R.P., Ledebt, A., Deconinck, F.J.A., Savelsbergh, G.J.P. The effects of a short proprioceptive training with (mirror) visual feedback in individuals with congenital hemiplegia. *New strategies to optimize the acquisition and consolidation of motor skills, a satellite symposium of the FENS forum of neuroscience.* Barcelona, Spain, 12-13 July 2012.

Smorenburg, A.R.P., Ledebt, A., Deconinck, F.J.A., Savelsbergh, G.J.P. Accuracy of unimanual vs. bimanual matching in individuals with Spastic Hemiparetic Cerebral Palsy. *Congress Mastery of Manual Skills 'Recent insights into typical and atypical development of manual ability'*. Groningen, The Netherlands, 19-21 April 2012.

Smorenburg, A.R.P., Ledebt, A., Deconinck, F.J.A., Savelsbergh, G.J.P. Visual feedback of the non-moving limb improves active joint-position sense of the impaired limb in Spastic Hemiparetic Cerebral Palsy. *VUMC day of science and technology.* Amsterdam, The Netherlands, 9 March 2012.

Smorenburg, A.R.P., Ledebt, A., Deconinck, F.J.A., Savelsbergh, G.J.P. Visual feedback of the non-moving limb improves active joint-position sense of the impaired limb in Spastic Hemiparetic Cerebral Palsy. *3rd Annual MOVE Research Meeting.* Amsterdam, The Netherlands, 28 September 2011.

Smorenburg, A.R.P., Ledebt, A., Deconinck, F.J.A., Savelsbergh, G.J.P. Visual feedback of the non-moving limb improves active joint-position sense of the impaired limb in Spastic Hemiparetic Cerebral Palsy. *21st Annual Meeting of the Society for the Neural Control of Movement*, San Juan, Puerto Rico, 26-30 April 2011.

Smorenburg, A.R.P., Ledebt, A., Deconinck, F.J.A., Savelsbergh, G.J.P. Joint-position sense of the impaired limb can be improved by visual feedback of the non-moving limb in children with Spastic Hemiparetic Cerebral Palsy. *VUMC day of science and technology*. Amsterdam, The Netherlands, 11 March 2011.

Smorenburg, **A.R.P.,** Ledebt, A., Deconinck, F.J.A., Savelsbergh, G.J.P. Children with Spastic Hemiparetic Cerebral Palsy show deficits in joint-position sense of the upper limbs. *VUMC day of science and technology.* Amsterdam, The Netherlands, 11 March 2011.

Smorenburg, A.R.P., Ledebt, A., Feltham, M.G., Deconinck, F.J.A., Savelsbergh, G.J.P. Visual feedback of the impaired limb provided by a mirror increases neuromuscular activity of the impaired limb in Spastic Hemiparetic Cerebral Palsy. *VUMC day of science and technology*. Amsterdam, The Netherlands, 11 March 2011.

Smorenburg, A.R.P., Ledebt, A., Deconinck, F.J.A., Savelsbergh, G.J.P. Children with Spastic Hemiparetic Cerebral Palsy show deficits in joint-position sense of the upper limbs. *Annual Research Conference Manchester Metropolitan University.* Manchester, United Kingdom, 10 December 2010. ISBN: 978-1-905476-54-1

Smorenburg, A.R.P., Ledebt, A., Deconinck, F.J.A., Savelsbergh, G.J.P. Joint-position sense of the impaired limb in Spastic Hemiparetic Cerebral Palsy can be influenced by visual feedback of the non-moving limb. *Annual Research Conference Manchester Metropolitan University.* Manchester, United Kingdom, 10 December 2010. ISBN: 978-1-905476-54-1

Smorenburg, A.R.P., Ledebt, A., Feltham, M.G., Deconinck, F.J.A., Savelsbergh, G.J.P. Visual feedback of the impaired arm provided by a mirror increases neuromuscular activity in Spastic Hemiparetic Cerebral Palsy. *Annual Research Conference Manchester Metropolitan University.* Manchester, United Kingdom, 10 December 2010. ISBN: 978-1-905476-54-1

Smorenburg, A.R.P., Ledebt, A., Deconinck, F.J.A., Savelsbergh, G.J.P. Joint-position sense of the impaired limb can be improved by visual feedback of the non-moving limb in children with Spastic Hemiparetic Cerebral Palsy. *2nd Annual MOVE Research Meeting 2010.* Amsterdam, Nederland, 24 September 2010.

Smorenburg, A.R.P., Ledebt, A., Deconinck, F.J.A., Savelsbergh,G.J.P. Proprioception in Spastic Hemiparetic Cerebral Palsy (preliminary results). *PhD day for Human Movement Sciences.* Amsterdam, The Netherlands, 11 June 2010.

Van Kampen, P.M., Ledebt, A., **Smorenburg, A.R.P.,** Vermeulen, J., van der Kamp, J., Savelsbergh, G.J.P. Action planning in children with Spastic Hemiparetic Cerebral Palsy: visual guidance during interception. *International Symposium: “A global status quo on Cerebral Palsy, with a view to the future”,* Utrecht, The Netherlands, 5-7 November 2009.

Oral presentations

Smorenburg, A.R.P. *“Proprioception and mirror visual feedback in children with hemiparetic cerebral palsy”.* Institute of Neuroscience and Pharmacology – Panum, University of Copenhagen, Denmark, 26 January 2012 (*invited talk).*

Smorenburg, A.R.P. *“The effects of mirror visual feedback in children with hemiparetic cerebral palsy”.* Helene Elsass Center Copenhagen, Denmark, 25 January 2012 (*invited talk).*

Smorenburg, A.R.P. De ‘mirror box’ illusie: van onderzoek naar therapie:
Wetenschappelijk onderzoek naar het effect van de ‘mirror box’ illusie op proprioceptie bij kinderen met Spastisch Hemiparetische Cerebrale Parese. Workshop on the annual conference of the Dutch Academy of Childhood Disability, 8 October 2011.

Appendix B

Abstracts of conference proceedings
Journal publications

Smorenburg, A.R.P., Ledebt, A., Deconinck, F.J.A., Savelsbergh, G.J.P. Accuracy of unimanual vs. bimanual matching in individuals with Spastic Hemiparetic Cerebral Palsy. *8th FENS forum of neuroscience.* Barcelona, Spain, 14-18 July 2012.

Kinematics of the impaired arm in children with Spastic Hemiparetic Cerebral Palsy (SHCP) improve during symmetrical bimanual movements. However, to the best of our knowledge no study examined the effect of symmetrical bimanual movements on movement accuracy. Movement accuracy can serve as a measure of proprioceptive accuracy, which has been shown previously to be deteriorated in SHCP-children. Therefore, the present study focused on movement accuracy in unimanual and bimanual symmetrical movements. Moreover, in the light of the positive reports about mirror therapy for treating arm dysfunction we also examined the effect of mirror visual feedback (MVF) of the less-impaired arm on movement accuracy of the impaired arm.

Participants with SHCP were asked to match the position of a target either with the impaired arm only (unimanual condition) or with both arms at the same time (bimanual condition). In both conditions a divide (opaque screen or mirror) in between the arms masked vision of the impaired arm. Matching accuracy was measured by the absolute difference between the impaired arm and the target at the end of the movement.

The results showed that absolute endpoint error was smaller in the bimanual compared to the unimanual condition, while there was no effect of MVF (i.e. similar error with screen and mirror). Inspection of individual data, however, showed that 13 out of 23 participants did experience a positive effect of MVF. Moreover, significant positive correlations seem to suggest that individuals with lower proprioceptive accuracy in the baseline condition (screen) seem to benefit more from MVF. This then suggests that MVF might be a valuable tool for rehabilitation but only for a subset of the individuals with SHCP. It is suggested that the large variability in response is caused by the heterogeneity in the CP population related to e.g. size and location of the lesion.

Smorenburg, A.R.P., Ledebt, A., Deconinck, F.J.A., Savelsbergh, G.J.P. The effects of a short proprioceptive training with (mirror) visual feedback in individuals with congenital hemiplegia. *New strategies to optimize the acquisition and consolidation of motor skills, a satellite symposium of the FENS forum of neuroscience.* Barcelona, Spain, 12-13 July 2012.

Objectives:

To examine the effects of a short proprioceptive training with (mirror) visual feedback in individuals with Spastic Hemiparetic Cerebral Palsy (SHCP).

Methods:

16 participants with SHCP (15.8 ± 2.5 years) performed a proprioceptive training which consisted of 40 bimanual symmetrical movements towards a visible target. A divide in between the arms occluded the impaired arm from view. To test the effect of mirror visual feedback, the divide was either an opaque screen or a mirror. For the screen-group, only the less-impaired arm was visible during training. For the mirror-group the less-impaired arm and its mirror reflection were visible. At the end of each trial, participants received feedback about the accuracy of the impaired arm.

A pretest was performed one week before training and the training was followed by a posttest (immediately after training) and a retention-test (1 week after training). Procedures of the pre-, post-, and retention-test were similar: participants were asked to match 5 target positions, scaled to the individual maximum reaching distance, with either the less-impaired arm only, the impaired arm only or both arms at the same time, without vision of either arm. The difference between the impaired arm and the target at the end of the movement (absolute error) was measured.

Results:

Movement accuracy of the impaired arm improved as a result of the training (error in posttest was smaller than error in pretest), but this improvement was similar for both training groups (i.e. mirror-group and screen-group). The error in the retention-test returned to pretest level indicating that the training was probably too short to see long-term effects. Finally, adding visual feedback of the less-impaired arm in the training immediately decreased the matching error of the impaired arm with respect to the pretest. This suggests that visual feedback of the less-impaired arm provides an important source of information to match the impaired arm accurately.

Conclusions:

Proprioceptive training with visual feedback of the less-impaired arm improves movement accuracy of the impaired arm in children and adolescents with congenital hemiplegia. However, mirror visual feedback did not have an additional (beneficial) effect on the training.

Smorenburg, A.R.P., Ledebt, A., Deconinck, F.J.A., Savelsbergh, G.J.P. Accuracy of unimanual vs. bimanual matching in individuals with Spastic Hemiparetic Cerebral Palsy. *Congress Mastery of Manual Skills 'Recent insights into typical and atypical development of manual ability'.* Groningen, The Netherlands, 19-21 April 2012.

Previous studies demonstrated that the kinematics of the impaired arm in children with Spastic Hemiparetic Cerebral Palsy (SHCP) improve during specific symmetrical bimanual movements (e.g. Steenbergen et al., 2006). However, to the best of our knowledge no study examined the effect of symmetrical bimanual movements on movement accuracy. Movement accuracy can serve as a measure of proprioceptive accuracy, which has been shown previously to be deteriorated in children with SHCP compared to typically developing peers (Smorenburg et al. in press). Therefore, the present study focused on the movement accuracy in unimanual and bimanual symmetrical goal-directed movements. Moreover, in the light of the positive reports about mirror therapy for treating arm dysfunction (e.g. Altschuler et al., 1999) we also examined the effect of mirror visual feedback of the less-impaired arm on the movement accuracy of the impaired arm.

23 participants with SHCP (mean age 14.2 ± 2.9; 5 females, 18 males) were asked to match the position of a target either with the impaired arm only (unimanual condition) or with both arms at the same time (bimanual condition). In both conditions a divide in between the arms masked the vision of the impaired arm. To test the effect of mirror visual feedback of the less-impaired arm on the matching accuracy, the divide was either an opaque screen or a mirror. For 4 target positions, scaled to the individual maximum reaching distance, the difference between the impaired arm and the target at the end of the movement (absolute error) was measured.

The results showed that the absolute endpoint error was smaller in the bimanual condition compared to the unimanual condition (3.1 vs. 4.3 cm), while there was no effect of mirror visual feedback (i.e. absolute error with opaque screen and mirror were similar). Inspection of the individual data, however, showed that 13 out of 23 participants did experience a positive effect of mirror visual feedback (error was smaller in the mirror compared to the screen condition). A significant positive correlation between the error in the screen condition and the difference score between the screen and the mirror condition further seems to suggest that individuals with lower proprioceptive accuracy in the baseline condition (screen) seem to benefit more from mirror visual feedback. This then suggests that mirror visual feedback might be a valuable tool for rehabilitation but only for a subset of the individuals with SHCP. It is thought that the large variability in response is caused by the heterogeneity in the CP population related to e.g. size and location of the lesion.

Smorenburg, A.R.P., Ledebt, A., Deconinck, F.J.A., Savelsbergh, G.J.P. Joint-position sense of the impaired limb can be improved by visual feedback of the non-moving limb in children with Spastic Hemiparetic Cerebral Palsy. *VUMC day of science and technology.* Amsterdam, The Netherlands, 9 March 2012.

Introduction

In this study we examined the effect of static visual feedback and static mirror visual feedback (i.e. a mirror image of the non-moving limb) on one component of proprioception, the joint-position sense (JPS) in children with Spastic Hemiparetic Cerebral Palsy (SHCP).

Methods

Participants were asked to match the position of one arm to that of the contra-lateral arm, using a device consisting of two moveable handles on a sagittal track. The task was performed in three conditions: 1) without visual feedback (no vision), 2) with visual feedback of the non-moving limb (screen), and 3) with visual feedback of the non-moving limb and its mirror reflection (mirror). The endpoint error, distance between the hands at the end of the movement, was used as indicator of JPS.

Results

JPS of the impaired limb was more accurate when static visual feedback was available compared to the no vision condition. However, static mirror visual feedback did not have any additional effect on JPS. These results indicate that children with SHCP can integrate static visual and proprioceptive feedback into an egocentric reference frame, which improves their JPS. Additionally, while static mirror visual feedback does not seem to add information to this reference frame, the mirror image did not lead to a sensory conflict.

Conclusions

Static visual feedback of the less-impaired limb improves the matching accuracy of the impaired limb in children with SHCP indicating that they are able to integrate visual and proprioceptive feedback into one egocentric reference frame. This provides possibilities for proprioceptive training.

Smorenburg, A.R.P., Ledebt, A., Deconinck, F.J.A., Savelsbergh, G.J.P. Visual feedback of the non-moving limb improves active joint-position sense of the impaired limb in Spastic Hemiparetic Cerebral Palsy. *3rd Annual MOVE Research Meeting.* Amsterdam, The Netherlands, 28 September 2011.

This study examined the active joint-position sense in children with Spastic Hemiparetic Cerebral Palsy (SHCP) and the effect of static visual feedback and static mirror visual feedback, of the non-moving limb, on the joint-position sense. Participants were asked to match the position of one upper limb with that of the contralateral limb. The task was performed in three visual conditions: without visual feedback (no vision); with visual feedback of the non-moving limb (screen); and with visual feedback of the non-moving limb and its mirror reflection (mirror). In addition to the proprioceptive measure, a functional test (Quality of Upper Extremity Skills Test [QUEST]) was performed and the amount of spasticity was determined in order to examine their relation with the proprioceptive ability. Results showed that the accuracy of matching was significantly influenced by the distance that had to be covered by the matching limb; a smaller distance resulted in smaller errors. Moreover it was demonstrated that static (mirror) visual feedback improved the matching accuracy. A clear relation between functionality, as measured by the QUEST, and active joint-position sense was not found. This might be explained by the availability of visual information during the performance of the QUEST. It is concluded that static visual feedback improves matching accuracy in children with SHCP and that the initial distance between the limbs is an influential factor which has to be taken into account when measuring joint-position sense.

Smorenburg, A.R.P., Ledebt, A., Deconinck, F.J.A., Savelsbergh, G.J.P. Visual feedback of the non-moving limb improves active joint-position sense of the impaired limb in Spastic Hemiparetic Cerebral Palsy. *Annual Meeting of the Society for the Neural Control of Movement*, San Juan, Puerto Rico, 26-30 April 2011.

This study examined the active joint-position sense in children with Spastic Hemiparetic Cerebral Palsy (SHCP) and the effect of static visual feedback and static mirror visual feedback, of the non-moving limb, on the joint-position sense. Participants were asked to match the position of one upper limb with that of the contralateral limb. The task was performed in three visual conditions: without visual feedback (no vision); with visual feedback of the non-moving limb (screen); and with visual feedback of the non-moving limb and its mirror reflection (mirror). In addition to the proprioceptive measure, a functional test [Quality of Upper Extremity Skills Test (QUEST)] was performed and the amount of spasticity was determined in order to examine their relation with the proprioceptive ability. Results showed that the accuracy of matching was significantly influenced by the distance that had to be covered by the matching limb; a smaller distance resulted in smaller errors. Moreover it was demonstrated that static (mirror) visual feedback improved the matching accuracy. A clear relation between functionality, as measured by the QUEST, and active joint-position sense was not found. This might be explained by the availability of visual information during the performance of the QUEST. It is concluded that static visual feedback improves matching accuracy in children with SHCP and that the initial distance between the limbs is an influential factor which has to be taken into account when measuring joint-position sense.

Smorenburg, A.R.P., Ledebt, A., Deconinck, F.J.A., Savelsbergh, G.J.P. Joint-position sense of the impaired limb can be improved by visual feedback of the non-moving limb in children with Spastic Hemiparetic Cerebral Palsy. *VUMC day of science and technology*. Amsterdam, The Netherlands, 11 March 2011.

Introduction

In this study we examined the effect of static visual feedback and static mirror visual feedback (i.e. a mirror image of the non-moving limb) on one component of proprioception, the joint-position sense (JPS) in children with Spastic Hemiparetic Cerebral Palsy (SHCP).

Methods

Participants were asked to match the position of one arm to that of the contra-lateral arm, using a device consisting of two moveable handles on a sagittal track. The task was performed in three conditions: 1) without visual feedback (no vision), 2) with visual feedback of the non-moving limb (screen), and 3) with visual feedback of the non-moving limb and its mirror reflection (mirror). The endpoint error, distance between the hands at the end of the movement, was used as indicator of JPS.

Results

JPS of the impaired limb was more accurate when static visual feedback was available compared to the no vision condition. However, static mirror visual feedback did not have any additional effect on JPS. These results indicate that children with SHCP can integrate static visual and proprioceptive feedback into an egocentric reference frame, which improves their JPS. Additionally, while static mirror visual feedback does not seem to add information to this reference frame, the mirror image did not lead to a sensory conflict.

Conclusions

Static visual feedback of the less-impaired limb improves the matching accuracy of the impaired limb in children with SHCP indicating that they are able to integrate visual and proprioceptive feedback into one egocentric reference frame. This provides possibilities for proprioceptive training.

Smorenburg, A.R.P., Ledebt, A., Deconinck, F.J.A., Savelsbergh, G.J.P. Children with Spastic Hemiparetic Cerebral Palsy show deficits in joint-position sense of the upper limbs. *VUMC day of science and technology.* Amsterdam, The Netherlands, 11 March 2011.

Introduction

Contradictory findings exist regarding the integrity of proprioception, i.e. sense of body parts in space, in individuals with Spastic Hemiparetic Cerebral Palsy (SHCP). Aim of this study was to get more insight into one component of proprioception, the joint-position sense (JPS) of the upper limbs in this population.

Methods

We compared the ability to match the position of one arm to that of the contra-lateral arm in a group of children with SHCP and typically developing (TD) children. The task was performed without visual information using a device consisting of two moveable handles on a sagittal track, with either the dominant or non-dominant limb as reference. The endpoint error, distance between the limbs at the end of the movement, was used as indicator of JPS.

Results

In the SHCP group, JPS of the impaired limb was worse than that of the less-impaired limb and both limbs showed less accurate JPS compared to the limbs of the TD group. Furthermore, larger errors were made when the distance to be covered was larger and this was more pronounced in the SHCP group.

Conclusions

These results show that in children with SHCP, JPS of both upper limbs is affected and therefore should be targeted in therapy. Moreover, the distance to be covered is suggested to take into account when determining joint-position sense.

Smorenburg, A.R.P., Ledebt, A., Feltham, M.G., Deconinck, F.J.A., Savelsbergh, G.J.P. Visual feedback of the impaired limb provided by a mirror increases neuromuscular activity of the impaired limb in Spastic Hemiparetic Cerebral Palsy. V*UMC day of science and technology*. Amsterdam, The Netherlands, 11 March 2011.

Introduction

When during a bimanual movement task, children with Spastic Hemiparetic Cerebral Palsy (SHCP) have access to mirror visual feedback (MFB) of their less-impaired limb, excessive interlimb variability and neuromuscular intensity in arm muscles decrease. Aim of the current study was to determine whether these positive effects were specific to the type of information provided by the mirror image or were generic responses to the visual illusion.

Methods

Children with SHCP were instructed to produce a symmetrical circular movement with both hands in two conditions: 1) with MFB of the less-impaired arm, 2) with MFB of the impaired arm. MFB was generated by means of a mirror that was placed in between the two arms, perpendicular to the chest.

Results

Seeing two impaired arms (the real arm and its mirror image) was found to increase the relative duration of eccentric muscle activity in the Biceps Brachii Brevis of the impaired arm whereas seeing two less-impaired arms decreased the relative duration of eccentric muscle activity of the Biceps.

Conclusions

This finding suggests that the positive effects of MFB are specific to the type of information provided and that the mechanism underpinning these effects possibly involves the elimination of a conflict between an internal motor representation and afferent feedback.

Smorenburg, A.R.P., Ledebt, A., Deconinck, F.J.A., Savelsbergh, G.J.P. Children with Spastic Hemiparetic Cerebral Palsy show deficits in joint-position sense of the upper limbs. *Annual Research Conference Manchester Metropolitan University.* Manchester, United Kingdom, 10 December 2010. ISBN: 978-1-905476-54-1

Spastic Hemiparetic Cerebral Palsy (SHCP) is a disorder characterized by spasticity on one side of the body. It is known that the impaired limb has a deteriorated functionality compared to the less-impaired limb. Nevertheless, contradictory findings exist with respect to the proprioception of the impaired and the less-impaired limb and the difference with both limbs of typically developing (TD) children. The current study aimed to get more insight into the joint-position sense of the upper limbs in children with SHCP and to compare this to the joint-position sense of TD children. Joint-position sense was showed to be influenced by the distance between the limbs at the start of the movement: larger distances resulted in bigger errors for both groups. However, this effect was more pronounced in the CP group. In addition, for the CP group the impaired limb showed an impaired joint-position sense compared to the less-impaired limb and to both limbs of the TD group. Furthermore, also the less-impaired limb showed a deteriorated joint-position sense compared to the dominant limb of the TD group. It is concluded that the joint-position sense of both upper limbs in children with SHCP is deteriorated compared to the upper limbs of TD children.

Smorenburg, A.R.P., Ledebt, A., Deconinck, F.J.A., Savelsbergh, G.J.P. Joint-position sense of the impaired limb in Spastic Hemiparetic Cerebral Palsy can be influenced by visual feedback of the non-moving limb. *Annual Research Conference Manchester Metropolitan University.* Manchester, United Kingdom, 10 December 2010.
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Active joint-positions sense is deteriorated in individuals with Spastic Hemiparetic Cerebral Palsy. Visual feedback of the moving upper limb (dynamic visual feedback) improves the joint-position sense of that same limb. However, visual feedback of the non-moving upper limb (static visual feedback) was showed not to be of influence in individuals with bilateral CP. The current study focused on the effect of static visual feedback on the joint-position sense in children with SHCP. In addition, the study examined the effect of mirror visual feedback of the non-moving upper limb on the joint-position sense. It was expected that the mirror would either provide an extended reference frame in which the moving hand could be slid, facilitating the contralateral matching or would create a conflict situation resulting in deteriorated ability to use joint-position sense. It was found that joint-position sense of the impaired limb improved in a condition in which static visual feedback was available compared to a condition without any visual feedback. It is suggested that the static visual feedback and the proprioceptive feedback of the non-moving upper limb are integrated into one egocentric reference frame which facilitates the contralateral matching. Nevertheless, mirror static visual feedback did not had any additional effect on the joint-position sense. This might be caused by a short exposure time, the discrete movement or the involvement of self-induced movement in the task.

Smorenburg, A.R.P., Ledebt, A., Feltham, M.G., Deconinck, F.J.A., Savelsbergh, G.J.P. Visual feedback of the impaired hand provided by a mirror increases neuromuscular activity in Spastic Hemiparetic Cerebral Palsy. *Annual Research Conference Manchester Metropolitan University.* Manchester, United Kingdom, 10 December 2010. ISBN: 978-1-905476-54-1

Mirror visual feedback has previously been showed to enhance the movement pattern of different patient groups like children with Spastic Hemiparetic Cerebral Palsy (SHCP). Mirror visual feedback of the less-impaired upper limb decreased the neuromuscular activity of the muscles in the impaired limb. In order to get more insight into the working mechanisms of the mirror, the current study investigated the effect of mirror visual feedback of the impaired upper limb on the neuromuscular activity of the muscles in the less-impaired limb. Seeing two impaired upper limbs in the mirror increased the neuromuscular activity of the muscles in the same limb whereas seeing two less-impaired limbs decreased the neuromuscular activity. This suggests that the type of visual feedback influences the neuromuscular activity and therefore creates opportunities for use in therapy.

Smorenburg, A.R.P., Ledebt, A., Deconinck, F.J.A., Savelsbergh, G.J.P. Joint-position sense of the impaired limb can be improved by visual feedback of the non-moving limb in children with Spastic Hemiparetic Cerebral Palsy. *2^{nd} Annual MOVE Research Meeting 2010.* Amsterdam, Nederland, 24 September 2010.

Individuals with Spastic Hemiparetic Cerebral Palsy are known to have a deteriorated proprioception in the impaired limb. The current study focused on the proprioception, or more specific joint-position sense, and the effect of static visual feedback (of the non-moving limb) and static mirror visual feedback on this joint-position sense. It was expected that static visual feedback could provide a reference frame for the moving limb and that static mirror visual feedback could provide either a reference frame in which the moving limb could be slid or could create a conflict situation which would deteriorate the performance. It was shown that static visual feedback indeed provided a reference frame since the performance improved compared to a no vision situation. However, it seems that the static mirror visual information is not used since the performance was equal to the static visual feedback situation. The effect of visual feedback was only visible for the impaired hand. In addition it was showed that distance between the hands influenced the active joint-position sense.

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Research in Developmental Disabilities

Matching accuracy in hemiparetic cerebral palsy during unimanual and bimanual movements with (mirror) visual feedback

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ABSTRACT

In the present study participants with Spastic Hemiparetic Cerebral Palsy (SHCP) were asked to match the position of a target either with the impaired arm only (unimanual condition) or with both arms at the same time (bimanual condition). The target was placed at 4 different locations scaled to the individual maximum reaching distance. To test the effect of mirror visual feedback of the less-impaired arm on the matching accuracy, an opaque screen or a mirror was placed in between the arms which masked vision of the impaired arm. Absolute endpoint error was smaller in the bimanual condition compared to the unimanual condition, but there was no effect of mirror visual feedback. Inspection of the individual data, however, showed that 13 out of 23 participants did experience a positive effect of mirror visual feedback. A positive correlation between the baseline error (screen) and the improvement in accuracy with mirror visual feedback seems to suggest that individuals with lower proprioceptive accuracy in the baseline condition may benefit more from mirror visual feedback. Together these findings indicate that bimanual therapy and therapy with mirror visual feedback might be valuable approaches for rehabilitation for a subset of the individuals with SHCP.

1. Introduction

Cerebral palsy (CP) is the most common paediatric physical disability (Stanley, Blair, & Alberman, 2000). The condition comprises a group of permanent disorders of movement and posture due to a lesion in the foetal or infant brain. In children with spastic hemiparetic cerebral palsy (SHCP), the motor impairments are mainly lateralized (i.e., one-sided) and the upper limb is usually more affected than the lower limb (Charles & Gordon, 2006; Humphreys, Whiting, & Pham, 2000). The brain damage in SHCP might also include areas that are involved in bimanual coordination such as the supplementary motor area (SMA) and areas in the parietal lobe (Serrien, Nirkko, Lovblad, & Wiesendanger, 2001; Serrien, Strens, Oliviero, & Brown, 2002; Steyvers et al., 2003). For this reason and because many daily activities require both hands, SHCP is often found to have a detrimental effect on bimanual tasks, and hence on many tasks of daily living (Gordon, 2011; Gordon & Steenbergen, 2008; Hung, Charles, & Gordon, 2004). Yet in tasks that typically require bimanual coordination using the non-dominant (impaired) hand is avoided and while they may become adept at using this compensatory strategy, this behaviour is considered to be inefficient and slow (Charles & Gordon, 2006; Gordon & Steenbergen, 2008). Interestingly though, there is evidence to suggest that the kinematics of the impaired arm are improved when the contralateral (less-impaired) arm

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performs an identical (symmetrical) action (Sugden & Utley, 1995; Utley & Sugden, 1998). These studies have mainly focused on kinematic variables (e.g., speed, trajectory or timing of the two limbs) and it remains to be determined whether accuracy of matching (of the impaired arm) is also favoured in a bimanual (symmetrical) condition. This will be the focus of our study.

Steenbergen, Hulstijn, de Vries and Berger (1996) studied the arm kinematics of young adolescents with SHCP during a reach-grasp-placement task. The participants were asked to pick up a ball and place it into a hole as quickly as possible with either one hand (one ball) or with two hands (two balls). It was found that the large differences in reaction time and total movement time between the hands in the unimanual condition decreased under bimanual conditions, indicating a tendency to move the impaired and less-impaired arm and hand in a symmetrical manner (interlimb coupling). Note though, that in this study the coupling was mainly unidirectional, i.e., the result of adaptations of the less-impaired hand to the movement of the impaired hand. Using similar reaching and grasping tasks Utley and Sugden (1998) further found that coupling (temporal and to a lesser extent also spatial) happened predominantly in the first part of the movement (and not in the grasping phase) and was facilitated when movements were performed under speeded conditions. However, in contrast to the findings of Steenbergen et al. (1996) the coupling was not unidirectional, i.e., temporal synchrony was the result of adaptations in both hands (see also Sugden & Utley, 1995). Finally, Volman (2005) demonstrated that interlimb coupling in children with SHCP is not just restricted to timing of the movement but also extends to spatial features. When children with hemiplegia were asked to draw a line with one hand and a circle with the other hand, the lines became more circular and the circles became more linear compared to a single handed condition. Neither the impaired nor the less-impaired arm dominated the coupling. Taken together, these findings demonstrate that even in individuals that have suffered unilateral brain damage that led to SHCP, typical bilateral neural interactions facilitating interlimb coupling seem to be present. This coupling appears to be dependent on a number of factors such as speed and the nature of the movement. It is however not known whether this coupling influences the accuracy of a matching action. Therefore, the first question that this study will address is: is the accuracy of matching with the impaired arm better when the less-impaired arm is moving towards the target simultaneously than when moving in isolation?

Matching accuracy can serve as a measure of proprioceptive accuracy, the sense of body parts in space, which is essential for movement performance. A previous study by Smorenburg, Ledebt, Deconinck and Savelsbergh (2012) has shown that children with SHCP perform poorer than their typically developing peers in a task where the position of one arm has to be matched with the other arm, which is indicative of deteriorated proprioceptive accuracy. If simultaneous movement of the less-impaired arm towards a target would improve the accuracy when matching with the impaired arm, this would support the integration of symmetric bimanual tasks in the training of impaired arm function.

A second phenomenon that has received a lot of attention with respect to the treatment of unilateral movement and pain disorders is mirror visual feedback (see Ramachandran & Altschuler, 2009, for a review). It is generated by placing a mirror between the upper limbs in the sagittal plane, so that one sees the real less- (or non-) impaired arm and its mirror reflection, which now is superimposed on the impaired arm. This creates the illusion of two hands moving in perfect symmetry. Mirror visual feedback has been demonstrated to alleviate (phantom) pain (McCabe et al., 2003; Ramachandran and Rogers-Ramachandran, 1996) and to improve movement performance in individuals with hemiparetic stroke (e.g., Altschuler et al., 1999; Stevens & Stoykov, 2003; Yavuzer et al., 2008). In addition, Feltham, Ledebt, Bennett, Deconinck, Verheul, and Savelsbergh (2010) suggested that mirror visual feedback might be a feasible therapeutic tool for children with SHCP. Performing a bimanual inward symmetrical movement with mirror visual feedback of the less-impaired arm decreased the variability of the interlimb coupling compared to a situation in which only the less-impaired arm was visible. Furthermore, in a subsequent study the authors showed that mirror visual feedback had favourable effects on the neuromuscular activity during a symmetric bimanual movement (Feltham, Ledebt, Deconinck, & Savelsbergh, 2010). The suggestions of Feltham, Ledebt, Bennett, et al. (2010), and Feltham, Ledebt, Deconinck, et al. (2010) were supported by a recently published study showing that 3 weeks of mirror therapy in children with SHCP resulted in improved grasp strength and upper limb dynamic position (Gygax, Schneider, & Newman, 2011). Smorenburg, Ledebt, Deconinck and Savelsbergh (2011), on the other hand, found that mirror visual feedback of the less-impaired arm did not influence endpoint accuracy of the impaired arm during unimanual matching. In this task the individuals were instructed to move the impaired limb to the position of the less-impaired limb, which was held passively at a target. In contrast to Feltham, Ledebt, Bennett, et al. (2010), Feltham, Ledebt, & Deconinck (2010) and Gygax et al. (2011) mirror visual feedback in the Smorenburg et al. study (2011) was 'static', i.e., the less-impaired arm was held at the target. This discrepancy in findings seems to suggest that mirror visual feedback might only be effective when both arms are intending to move symmetrically, which is a pertinent issue that needs to be clarified before therapy with mirror visual feedback can actually be integrated in the treatment of SHCP. Therefore, the current study will examine if mirror visual feedback might have a positive effect on the endpoint accuracy of a matching task (a measure of proprioceptive acuity) when the less-impaired arm is moving simultaneously with the impaired arm (symmetric bimanual movement), and thus when the mirror visual feedback is dynamic.

2. Methods

2.1. Participants

Twenty-five individuals with SHCP took part in the study, but 23 participants were included for analysis (14.2 ± 2.9 years, 5 females). All participants were recruited through the Dutch society for people with a physical handicap and their parents (BOSK)

Table 1

Participant characteristics. For each participant (P) the age in years, sex and impaired arm are indicated. In addition, the Tardieu scale for spasticity, the WeeFIM score and MACS level are mentioned. In the last two columns the aethiology of the disorder and the maximum reaching distances (MRD) of the impaired and less-impaired arm are given. The 'total' row provides the *M* and (SD) for age; total number of (fe)males; total number of left and right impaired arms; *M* and (SD) for Tardieu, WeeFIM, MACS and MRD of the (less-)impaired arm.

P	Age (years)	Sex	Impaired arm	Tardieu[a]	WeeFIM/MACS	Aethiology	MRD imp/less-imp (cm)[b]
1	11.1	M	Left	1.5	91/2	Unknown	35.5/38
2	14.8	M	Left	2	62/3	Schizencephaly right	33/36
3	13.7	M	Left	2	78/3	O_2 shortage during birth	33/40
4	14.0	M	Left	2	91/2	Cerebral infarction	31.5/33.7
5	13.3	M	Left	2	70/2	Premature (twins)	29/32
6	13.8	F	Left	1.5	91/2	O_2 shortage (twins)	27.3/29.5
7	13.0	M	Left	1	91/2	Hydrocephalus	20/24
8	14.5	M	Left	1	91/2	Stroke	30/31
9	14.6	M	Left	1	59/3	Streptococcen infection at 5 weeks	24/40
10	17.8	M	Left	1.5	90/1	Perinatal cerebral infarction	38/39
11	17.0	M	Left	1	91/1	Premature	25/29
12	18.7	M	Left	0.5	88/2	Cerebral infarction	25.5/29
13	9.6	M	Right	1	91/1	Cerebral infarction	34.5/35.5
14	14.7	M	Right	2	71/3	Unknown	33/38
15	12.8	M	Right	2	59/3	Cerebral infarction	26.5/38
16	9.3	F	Right	2	85/2	Hydrocephalus	30/33.3
17	16.2	M	Right	2	76/1	Unknown	40/40
18	12.7	F	Right	1	91/3	Thalamus infarction at birth	30/32.5
19	18.7	M	Right	1.5	91/3	Cerebral infarction	33/39
20	7.9	F	Right	1	91/1	Feverish convulsion	25/26
21	17.2	M	Right	Unknown	89/3	Cerebral infarction	22/28.5
22	17.7	F	Right	1.5	91/2	Stroke	22/29
23	14.5	M	Right	0.5	91/2	Premature	25/27
Total	14.2 (2.9)	5 F; 18 M	12L; 11 R.	1.4 (0.5)	83.4 (11.4)/2.1 (0.8)		29.3 (5.3)/33.4 (5.0)

[a] Tardieu scale for spasticity = mean of the individual scores for the biceps and the triceps.

[b] MRD = maximum reaching distance in cm for the impaired and the less-impaired arm.

and the Werkenrode school in Groesbeek (The Netherlands), a special education school. Two participants were not included for analysis; one participant was not able to finish the experiment due to fatigue, and another participant had absolute error values that were more than 2 standard deviations of the mean. The participants did not have a visual impairment (which was not corrected to normal), hearing impairment, pain in either of the upper limbs, visual neglect, Botox treatment in the past six months preceding the measurement, or any other neuromuscular disorder than SHCP. Moreover, participants were required to understand basic instructions in order to perform the measurement. Table 1 represents the participant characteristics. For each participant the level of spasticity was determined with the Tardieu scale which ranges from 0 to 3, with a higher score indicating higher levels of spasticity. Individual scores were obtained for the biceps brachii brevis and triceps brachii longus and combined into one total score. Functional independence in daily life, taking into account caregiver assistance and the use of special equipment, was measured with the motor items of the Functional Independence Measure for children (WeeFIM). The participant's parents filled in the WeeFIM questionnaire. WeeFIM scores can range from 1 to 91, with a higher score representing a better functional independence. Finally, the Manual Ability Classification System (MACS) describes how children use their hands during object handling and the degree of required assistance (Eliasson et al., 2006). The severity of performance and the degree of required assistance increases from MACS level 1 to 4. For more detailed information about the Tardieu, WeeFIM and MACS we refer to Appendix A.

Prior to testing, the participant's parents provided written informed consent. All procedures were approved by the institutional research ethics committee and in accordance with the Declaration of Helsinki.

2.2. Materials and procedures

The participant was seated on a height adjustable chair at a height adjustable table with the knees flexed to 90°. On the table a custom made wooden construction was placed which consisted of two handles on two separate parallel tracks 20 cm apart (see Fig. 1). The participant grasped the two handles (one in each hand), which could be moved in the anterior–posterior direction. The children were positioned such that the centre of the body was located in between the two tracks, with the beginning of the track 15 cm from the trunk. The position of the handles was recorded outside the wooden construction using one Optotrak unit with three infrared cameras (3020 Optotrak, Northern Digital Inc., Waterloo, Canada) at a sample rate of 200 Hz. A mirror or opaque screen, which was placed in between the tracks and perpendicular to the chest, served to elicit mirror visual feedback of the less-impaired arm or visual feedback of the less-impaired arm only.

Before the start of the measurement, the maximum reaching distance was determined (MRD). The child was asked to grasp the handles and extent the elbows as far a possible without bending the trunk forward. The MRD of the impaired arm

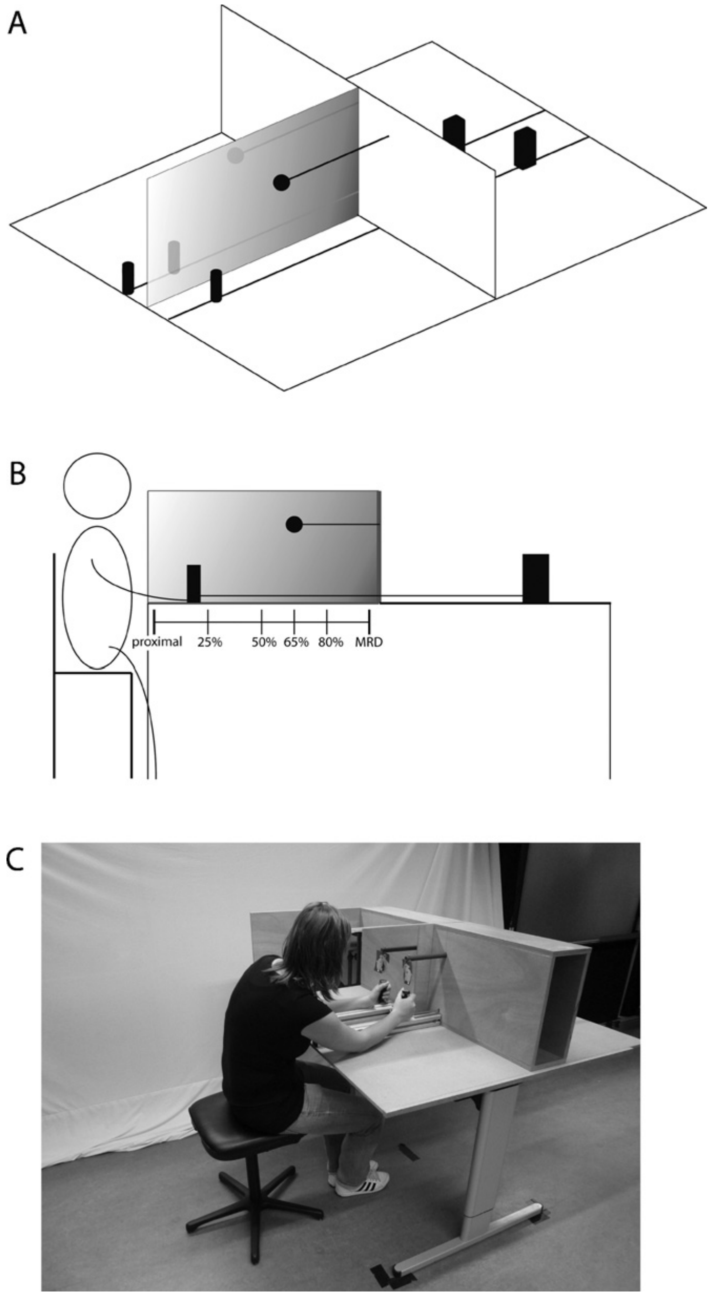

Fig. 1. (A) Top view of the experimental setup with the two handles that could be moved back and forth along the track. The divide between the arms was either an opaque screen or a mirror. The position of the handles outside the box was measured with an Optotrak camera (not depicted here). (B) Side view of the experimental setup. The proximal starting position and the four target positions (25% MRD, 50% MRD, 65% MRD, 80% MRD) are indicated. Note that the target positions were determined based on the maximum reaching distance of each child and thus differed per participant. (C) Real-life picture of the experimental setup.

was used to calculate the different target positions for the matching task. If a participant was unable to grip the handle due to physical impairment, the experimenter placed the hand on top of the handle. Each participant performed two tasks: a unimanual matching task and a bimanual matching task. The order of the tasks was randomly assigned to the participants. In the following paragraphs the procedures for the unimanual and the bimanual matching task will be explained.

2.2.1. *Unimanual matching task*

In the unimanual matching task, a target was placed at 25%, 50%, 65%, or 80% of the MRD on the side of the less-impaired hand. The less-impaired hand was placed on the lap and the impaired hand was holding the handle on the other side of the mirror/screen and was not visible. The participant was asked to match the position of the target by actively moving the impaired arm (the impaired hand always started proximal to the body at the start of the track, i.e., 0% MRD). The task was performed in two different visual conditions: a screen condition in which only the target was visible and a mirror condition in which the target and its mirror reflection were visible. Each combination of visual condition (2) and target position (4) was performed twice, which resulted in 16 trials. The order of the visual condition and the target positions were randomly assigned to the participants.

2.2.2. Bimanual matching task

In the bimanual matching task a target was placed at 25%, 50%, 65%, or 80% of the MRD on the side of the less-impaired arm. The participant was asked to match the target position with both hands, i.e., to move both hands towards the target as symmetrically as possible starting with the handles at the beginning of the track, i.e., 0% MRD. Similar to the unimanual task, the bimanual task was performed in two different visual conditions: a screen condition in which the target and the (moving) less-impaired arm could be seen and a mirror condition in which the participant saw the target, the less-impaired arm and its mirror reflection.

Each combination of visual condition (2) and target position (4) was performed twice (16 trials in total) and the order of the visual condition and the target positions were randomly assigned to the participants.

2.3. Data analysis

Custom-made Matlab programmes (The Mathworks, version 7.1) were used to analyze the kinematics and matching accuracy (absolute error) of the movement. The start of the movement was defined as the moment at which the movement velocity rose above 5 mm/s for the first time and the hand was moving in a forward direction. The end of the movement was defined as the moment at which the velocity finally fell below 5 mm/s (van Roon, Steenbergen, & Meulenbroek, 2005). Absolute error was determined as the difference in cm between the target and the impaired arm at the end of the movement. In addition, we calculated average movement velocity (cm/s; total distance covered divided by total movement time) and relative movement smoothness. Relative movement smoothness was defined as the number of peaks in the velocity plot of the entire movement divided by the total distance covered during each movement. The number of peaks was determined by searching the velocity curve for local minima and maxima. An increase in velocity between an adjacent minimum and maximum that exceeded the threshold value (10% of the maximum velocity) was counted as a peak (Chang, Wu, Wu, & Su, 2005; Kamper, McKenna-Cole, Kahn, & Reinkensmeyer, 2002).

2.4. Statistical analysis

In order to examine differences in absolute error, mean velocity and movement smoothness of the impaired arm between the unimanual and bimanual task and to examine the effects of visual feedback and target distance on these variables, a 3-way ANOVA was performed with repeated measures on the factors Task (unimanual, bimanual), Visual condition (mirror, screen), and Distance (25%, 50%, 65%, 80% MRD).

In addition, for the bimanual task differences in kinematics between the impaired and the less-impaired arm and the effect of Visual condition and Distance were investigated with a 3-way repeated measures ANOVA with Arm (impaired, less-impaired), Visual condition (mirror, screen), and Distance (25%, 50%, 65%, 80% MRD) as within factors.

The significance level was set at 0.05. In case sphericity assumptions were violated, Greenhouse-Geisser adjustments were made.

3. Results

All 23 participants were able to complete the experiment according to the instructions and all participants could perform a bimanual symmetrical movement as indicated by the small differences in starting time between the arms (difference between arms in mirror condition: $M = -0.05$ s, $SD = 0.25$, $t_{22} = -0.96$, $p = 0.035$; difference between arms in screen condition: $M = 0.04$ s, $SD = 0.28$, $t_{22} = 0.74$, $p = 0.47$). Although slightly larger, the differences in end time between the arms were also relatively small (difference between arms in mirror condition: $M = 0.48$ s, $SD = 1.19$, $t_{22} = 1.95$, $p = 0.06$; difference between arms in screen condition: $M = 0.59$ s, $SD = 0.94$, $t_{22} = 3.04$, $p = 0.006$).

Nevertheless, one trial was excluded because participant 15 did not perform a symmetrical bimanual movement, i.e., the movement of the impaired hand was initiated after the movement of the less-impaired arm was finished. In addition, 14 out of 368 trials in the bimanual condition had to be excluded from the analysis [PP 1 (2 trials), 3 (4), 15 (3), 8 (2), 23 (2), 12 (1)] because the less-impaired arm was not on the target location at the end of the movement. In case the difference between less-impaired arm and target was more than half of the distance between two consecutive target locations, the trials could not be assigned to either target distance and therefore they were excluded from analysis. This exclusion of trials meant that for some participants the value for a certain condition was based on one trial instead of the mean of two trials.

3.1. Unimanual vs. bimanual task (impaired arm)

3.1.1. Matching accuracy

Matching accuracy differed significantly between the unimanual and the bimanual task, and a significant Task by Distance effect indicated that this difference was distance dependent ($F_{3,66} = 3.16$, $p = 0.03$, *partial* $\eta^2 = 0.13$; see Fig. 2). Absolute error was smaller in the bimanual task compared with the unimanual task for all but the 25% MRD target position.

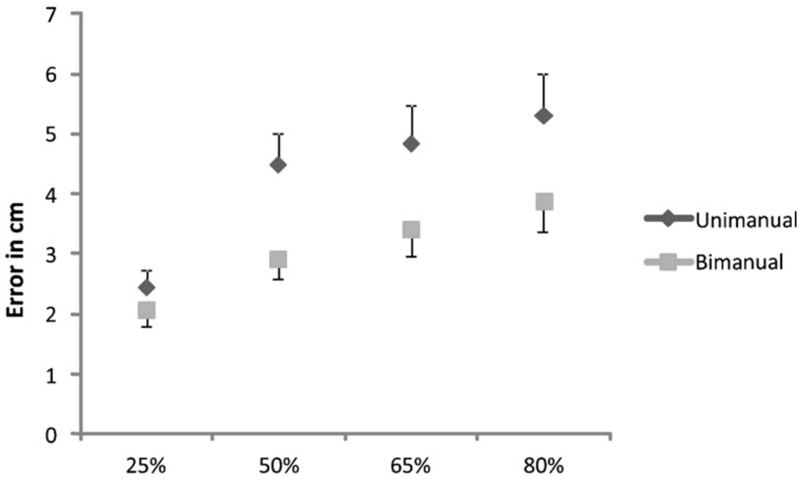

Fig. 2. The absolute error in cm (mean and SE) increased with increasing distance (25%, 50%, 65%, 80% MRD on the horizontal axis) for both the unimanual (dark grey) and the bimanual task (light grey).

In addition, absolute error was found to increase with increasing distance for both the unimanual and the bimanual task. However, between 50% and 65% and between 65% and 80% MRD the increase in error was not significant for either task.

Despite the significant Task-effect (unimanual vs. bimanual) on matching accuracy at group level, close inspection of the individual data showed that the advantage of moving simultaneously with the two hands was not present in all participants. In 14 out of 23 individuals absolute error in the bimanual condition was smaller than in the unimanual condition for 3 or 4 of the 4 target distances (see Table 2; *Bi*+ group). However, for both the *Bi*+ and the *Bi*− group it was demonstrated that the absolute error in the unimanual condition was positively correlated with the size of the decrease in error in the bimanual condition (Table 3), i.e., a larger error in the unimanual condition was related to a greater improvement in the bimanual condition.

Furthermore, as the repeated measures ANOVA showed, there was no effect of Visual condition on matching accuracy of the bimanual task (i.e., no interaction effect between Visual condition and Task), thus mirror visual feedback of the target did not affect absolute error. Inspection of the individual data of the bimanual task, however, indicated that in 13 out of 23 participants absolute error was smaller in the mirror condition compared to the screen for 3 or 4 of the 4

Table 2
Classification of the participants into groups. For each participant and each target distance (20%, 50%, 65%, 80% MRD) an asterisk (*) indicates when the error was smaller in the bimanual condition compared to the unimanual condition (left part of the table) and when the error was smaller in the mirror compared to the screen condition in the bimanual condition only (right part of the table). When in 3 or 4 out of 4 distances the error was smaller in the bimanual condition, the participant was assigned as performing better in the bimanual condition compared to the unimanual condition. For the screen/mirror comparison the same principle was used. When the error was smaller in the mirror condition compared to the screen condition (indicated with *) the participant was assigned to the *Mirror*+ group (i.e., *Mirror*+ = +).

Participant	*Bi*+ vs. *Bi*−					*Mirror*+ vs. *Mirror*−				
	25%	50%	65%	80%	*Bi*+ or *Bi*−?	25%	50%	65%	80%	*Mirror*+ or *Mirror*−?
1	*	*		*	+	*	*	*	*	+
2	*			*	–	*	*	*	*	+
3		*	*	*	+			*	*	–
4	*	*	*		+	*	*		*	+
5		*		*	–	*		*	*	+
6	*	*	*	*	+	*				–
7	*	*	*	*	+	*		*	*	+
8	*	*	*	*	+	*	*	*	*	+
9					–	*				–
10	*	*	*	*	+	*				–
11			*	*	–		*			–
12		*	*	*	+					–
13	*	*	*	*	+	*		*		–
14		*			–		*	*	*	+
15		*		*	–		*	*	*	+
16	*	*	*	*	+	*	*	*	*	+
17	*	*	*	*	+	*		*	*	+
18	*	*	*	*	+	*	*	*	*	+
19	*	*	*	*	+		*	*	*	+
20	*	*	*	*	+	*	*		*	+
21		*			–			*		–
22			*	*	–		*			–
23		*	*		–			*		–

Table 3
For each distance the correlations are reported between the error in the unimanual task (U25, U50, U65, U80) and the difference in error between the unimanual and the bimanual condition, i.e., error in the unimanual condition minus the error in the bimanual condition (DifUB25, DifUB50, DifUB65, DifUB80) for the *Bi*+ and the *Bi*− group. The table shows Pearson's *r* value and the corresponding *p*-value. Significant correlations are indicated with an asterisk.

Group	Correlation	Pearson *r*	*p*-Value
Bi+ ($n = 14$)	U25 vs. DifUB25	0.51	0.16
	U50 vs. DifUB50	0.98	<0.001*
	U65 vs. DifUB65	0.73	0.03*
	U80 vs. DifUB80	0.61	0.08
Bi− ($n = 9$)	U25 vs. DifUB25	0.36	0.21
	U50 vs. DifUB50	0.74	0.002*
	U65 vs. DifUB65	0.72	0.003*
	U80 vs. DifUB80	0.76	0.002*

distances (see Table 2; *Mirror+ group*). In Fig. 3 the mean errors in the screen and the mirror condition are depicted for the *Mirror*+ and the *Mirror*− group.

In order to reveal whether this variability in response to mirror visual feedback was related to the proprioceptive accuracy of the impaired arm when no mirror visual feedback was available, we examined for both groups (*Mirror*+ and *Mirror*−) the correlation between the error in the screen condition ('baseline condition') and the improvement in accuracy due to the mirror, i.e., the difference in error between the screen and the mirror condition. Table 4 shows these correlations and the corresponding *p*-values for the *Mirror*+ and the *Mirror*− group. No significant correlations were found for the *Mirror*− group, whereas significant positive correlations between the baseline error and the improvement in accuracy due to the mirror were observed for the *Mirror*+ group on all four distances. This suggests that for individuals who do better in the mirror than in the screen condition in the majority of the target distances (*Mirror*+ group), a larger error in the screen condition is related to a larger decrease in error in the mirror condition, i.e., to a higher degree of improvement in the mirror condition.

In addition, we examined with a Mann–Whitney *U* test whether the *Mirror*+ *and Mirror*− group differed in terms of scores on the MACS, WeeFIM and Tardieu scale. No differences between the groups were found for the MACS ($z = -0.69$, $p = 0.52$; mean rank *Mirror*+ = 12.81, *Mirror*− = 10.95) and the WeeFIM ($z = -0.40$, $p = 0.74$; mean rank *Mirror*+ = 11.54, *Mirror*− = 12.60). However, the *Mirror*+ group showed a higher average Tardieu score when compared to the *Mirror*− group (1.65 and 1.17, respectively; $z = -2.17$, $p = 0.04$; mean rank *Mirror*+ = 13.88, *Mirror*− = 8.06).

3.1.2. Average velocity

There was no effect of Task on average velocity ($F_{1,21} = 0.45$, $p = 0.51$, *partial* $\eta^2 = 0.02$; unimanual = 5.1 cm/s, bimanual = 4.8 cm/s). Moreover, Visual condition did not have an effect on the average velocity ($F_{1,21} = 1.25$, $p = 0.28$, *partial* $\eta^2 = 0.06$; *Mirror*: 4.7 cm/s; screen: 5.2 cm/s). However, a significant main effect of Distance was found ($F_{1.76,38.61} = 30.40$, $p < 0.001$, *partial* $\eta^2 = 0.58$), indicating an increase in velocity with increasing distance (25%: 3.50 ± 0.32; 50%: 4.83 ± 0.47; 65%: 5.45 ± 0.59; 80%: 5.99 ± 0.52).

3.1.3. Movement smoothness

A main effect of Distance ($F_{2.33,51.26} = 57.03$, $p < 0.001$, *partial* $\eta^2 = 0.72$) and a significant interaction effect between Task and Distance were found ($F_{1.92,42.27} = 60.21$, $p = 0.005$, *partial* $\eta^2 = 0.22$) for movement smoothness. No differences between

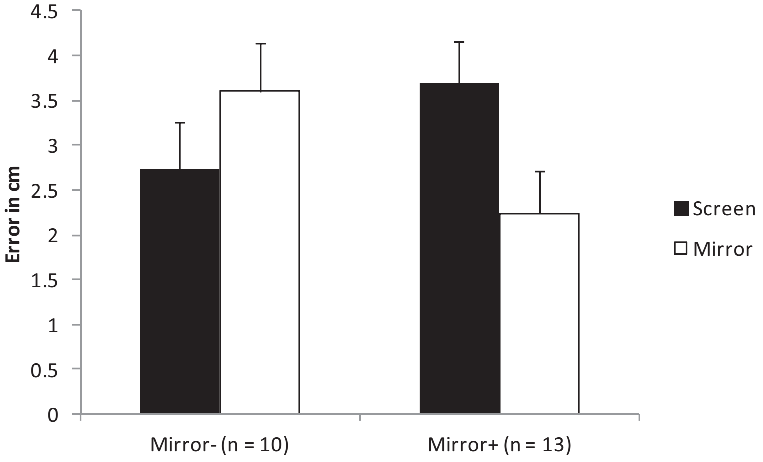

Fig. 3. Absolute error for the *Mirror*+ and the *Mirror*− group in the screen and the mirror condition. The Wilcoxon signed rank test revealed a significantly higher error in the screen compared to the mirror condition for the *Mirror*+ group ($z = -3.18$, $p < 0.00$). For the *Mirror*− group the error was higher in the screen compared to the mirror condition ($z = -2.50$, $p = 0.01$).

Table 4

For each distance the correlations are reported between the error in the screen condition (S25, S50, S65, S80) and the difference in error between the mirror and the screen condition, i.e., error in screen condition minus the error in mirror condition (DifMS25, DifMS50, DifMS65, DifMS80) for the *Mirror+* and the *Mirror−* group. The table shows the Pearson's *r* value and the corresponding *p*-value. Significant correlations are indicated with an asterisk.

Group	Correlation	Pearson *r*	*p*-Value
Mirror+ ($n = 13$)	S25 vs. DifMS25	0.69	0.009*
	S50 vs. DifMS50	0.76	0.002*
	S65 vs. DifMS65	0.70	0.007*
	S80 vs. DifMS80	0.69	0.009*
Mirror− ($n = 10$)	S25 vs. DifMS25	0.13	0.73
	S50 vs. DifMS50	−0.007	0.99
	S65 vs. DifMS65	0.31	0.39
	S80 vs. DifMS80	−0.43	0.22

the unimanual and the bimanual task were found on all of the four distances. However, for both the unimanual and the bimanual task the relative number of velocity peaks decreased (i.e., movement smoothness increased) with increasing distance (except for the unimanual task between 50% and 65% MRD and for the bimanual task between 65% and 80% MRD).

3.2. Bimanual task

In order to examine differences in kinematics between the impaired and the less-impaired arm, a repeated measures ANOVA was performed with Visual condition (mirror, screen), Distance (25%, 50%, 65%, 80% MRD) and Arm (impaired, less-impaired) as within factors. Moreover, in order to examine differences between the *Mirror+* and the *Mirror−* group this factor (*Mirror−* group) was included as between factor in the 3-way repeated measures ANOVA.

3.2.1. Average velocity

The ANOVA revealed a significant main effect of Visual condition ($F_{1,21} = 5.84$, $p = 0.03$, *partial* $\eta^2 = 0.22$). The average velocity was 0.7 cm/s lower in the mirror condition (4.6 ± 0.5 cm/s) compared to the screen condition (5.3 ± 0.7 cm/s). Furthermore, the significant main effects of Arm ($F_{1,21} = 5.14$, $p = 0.03$, *partial* $\eta^2 = 0.20$) and Distance ($F_{1.95,41.03} = 21.22$, $p < 0.001$, *partial* $\eta^2 = 0.50$) were modified by a significant interaction effect between Arm and Distance ($F_{2.57,53.90} = 9.62$, $p < 0.001$, *partial* $\eta^2 = 0.31$) and a significant interaction between Arm, Distance, and Mirror group ($F_{3,63} = 3.16$, $p = 0.03$, *partial* $\eta^2 = 0.13$; see Table 5).

Inspection of the 3-way interaction showed no differences between the *Mirror+* and the *Mirror−* group. For both the *Mirror+* group and the *Mirror−* group and both arms a significant increase in average velocity was found when the distance that had to be covered increased. Moreover, comparing the average velocity between the impaired and the less-impaired arm showed for both groups higher velocities in the less-impaired than in the impaired arm, but only for larger distances (65% and 80% MRD).

3.2.2. Movement smoothness

The analysis of the relative movement smoothness revealed a significant effect of Distance ($F_{1.86,38.97} = 33.96$, $p < 0.001$, *partial* $\eta^2 = 0.62$) and a significant interaction effect between Arm and Distance ($F_{1.71,36.00} = 3.76$, $p = 0.04$, *partial* $\eta^2 = 0.15$). For both the impaired and the less-impaired arm the relative number of velocity peaks decreased (i.e., movement smoothness increased) with increasing distance (except for the 65–80% MRD). In addition, the number of velocity peaks was higher in the impaired compared to the less-impaired arm, indicating a lower relative movement smoothness for the impaired arm, but only for the 80% MRD (impaired arm = 0.32 peaks/cm vs. less-impaired arm = 0.25 peaks/cm).

Table 5

Mean and standard error for the $V_{average}$ and Movement Smoothness. Values are given for each distance (25%, 50%, 65%, 80% MRD) in the unimanual and bimanual movement condition for the impaired and the less-impaired arm. Note that no values are reported for the less-impaired arm in the unimanual condition because this task was not performed in the present study.

		Unimanual	Bimanual	
	Distance	Impaired arm	Impaired arm	Less-impaired arm
$V_{average}$ (cm/s)	25%	3.71 ± 0.40	3.19 ± 0.40	3.16 ± 0.34
	50%	4.88 ± 0.52	4.79 ± 0.56	5.20 ± 0.59
	65%	5.54 ± 0.57	5.36 ± 0.73	6.08 ± 0.82
	80%	6.19 ± 0.56	5.80 ± 0.63	6.40 ± 0.64
Movement smoothness (peaks/cm)	25%	0.68 ± 0.069	0.51 ± 0.074	0.57 ± 0.088
	50%	0.43 ± 0.048	0.39 ± 0.071	0.38 ± 0.073
	65%	0.37 ± 0.044	0.33 ± 0.049	0.28 ± 0.054
	80%	0.30 ± 0.042	0.32 ± 0.071	0.25 ± 0.051

4. Discussion

This study examined the difference in matching accuracy of the impaired hand between a unimanual and a bimanual condition and the effects of mirror visual feedback on matching accuracy in children and adolescents with SHCP. Consistent with earlier studies that showed beneficial effects on the timing and the control of the impaired hand and arm when moving the two hands simultaneously (e.g., Steenbergen et al., 1996; Sugden & Utley, 1995; Utley & Sugden, 1998), we found a significant decrease in matching accuracy (37.5% on average) in the bimanual condition compared to the unimanual condition. In addition, mirror visual feedback led to better matching in 13 out of 23 participants. Together, these findings support the application of bimanual symmetrical movements and the use of mirror visual feedback in the treatment of upper limb function, though additional research is warranted to determine under what circumstances and for whom this approach is effective.

The underlying mechanism of the improved matching accuracy in the bimanual condition is probably related to facilitative processes resulting from bilateral connections throughout the central nervous system. For example, neural crosstalk is suggested to constrain homologous muscle groups to act as a single coordinative structure during bimanual symmetrical movements, which enhances the coupling between the limbs and also more abstract parameters (e.g., amplitude, force, direction; Cattaert, Semjen, & Summers, 1999; Swinnen & Wenderoth, 2004). In addition, we suggest that in the present study congruent visual and proprioceptive information of the less-impaired arm, which was available in the bimanual condition and presumably served as a frame of reference, may have facilitated accurate placement of the impaired arm (see also Smorenburg et al., 2011).

Consistent with other research (Smorenburg et al., 2011, 2012; van Beers, Sittig, & Denier van der Gon, 1998), larger errors were made in (unimanual and bimanual) matching movements with larger amplitude. Note that larger movements were also relatively faster and smoother. This counterintuitive finding for this population suffering from spasticity may be explained by the rather slow overall speed of movement execution. Spastic movement disruptions are commonly observed at higher speeds, and in this self-paced task it is likely that participants avoided detrimental effects of spasticity.

Concentrating on the effects of mirror visual feedback, the results of the present study showed that both hands moved slower in the mirror condition compared to the screen condition. Further, there was no improvement in accuracy of the impaired hand when mirror visual feedback of the less-impaired hand was available. Remarkably though, inspection of individual data revealed a positive effect of mirror visual feedback on matching accuracy in a considerable number of individuals (13 out of 23). In fact, mirror visual feedback seemed to hamper accurate placement of the impaired arm in the remainder of the group, which may explain the absence of a statistical effect at group level.

Explaining the mechanisms underlying the positive effect of the mirror remains speculative, but using transcranial magnetic stimulation (TMS) and advanced brain imaging techniques in healthy individuals, researchers have begun to uncover the neural basis of the mirror effects. For example, Garry, Loftus, and Summers (2005) have shown that the excitability of the ipsilateral[1] primary motor cortex (M1) is facilitated when healthy adults were viewing a mirror reflection of the moving hand (see also Nojima et al., 2012; Tominaga et al., 2011). In addition, mirror visual feedback was found to alter touch perception by enhancing the tactile sensitivity in the ipsilateral posterior parietal cortex (PPC; Ro, Wallace, Hagedorn, Farne, & Pienkos, 2004) and, further, to lead to increased activation within the ipsilateral superior temporal gyrus (STG; Matthys et al., 2009). Finally, the findings of Hamzei et al. (2012) suggest a remodelling of the motor system with a pivotal role for the contralateral[2] sensorimotor cortex (SMC) after training with the mirror (see also Michielsen et al., 2011). Apparently, mirror visual feedback has the capacity to induce plastic changes in brain regions directly involved in motor control (M1, SMC) and regions that have been linked with the mirror neuron system (PPC, STG).

The involvement of (part of) these specific regions might also (partly) explain the variability in response to mirror visual feedback across individuals. Staudt et al. (2002) found that the SHCP population may be functionally classified on the basis of the size of the lesion. Larger lesions are accompanied with a cortical reorganization of the primary motor cortex and premotor areas towards the contralesional cortex, whereas no reorganization is observed when the lesion is small. Wilke et al. (2009) on the other hand, found that the primary sensory cortex was preserved in the contralateral, lesioned hemisphere, irrespective of the extent of the lesion, which means that the sensorimotor control loop is disrupted when motor areas are relocated to the contralesional side. This variety in clinical picture might then be related to the variability in behavioural response to mirror visual feedback found in the current study. The idea that heterogeneity in patient groups, and more in particular variance in the neural resources, can explain the varying success of interventions is consistent with earlier findings in individuals with SHCP or a hemiparesis after stroke (McCombe Waller & Whitall, 2008; Ramachandran & Altschuler, 2009).

Our findings highlight that it is essential to determine which children might benefit most from therapy with mirror visual feedback, e.g., by using data on the side of the lesion or corticospinal reorganization. Unfortunately, lack of brain imaging and other neurophysiological data do not allow us to identify in which particular groups of children and adolescents mirror visual feedback may be favourable. However, behavioural evidence indicates that the extent of improvement in the mirror condition is related to the size of the error in baseline conditions. A similar result was found for the improvement under

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[1] Ipsilateral refers to the hemisphere at the same side of the moving arm which was visible in the mirror.

[2] Contralateral refers to the hemisphere contralateral to the moving arm which was visible in the mirror.

bimanual conditions, i.e., the improvement was larger when the error in the unimanual condition was greater. Moreover, a higher level of spasticity seemed to be related to a larger improvement in the mirror condition. This might suggest that both bimanual practice and practice with the mirror would be more effective in individuals with more severe problems of position sense. On the other hand, it is possible that the children, who did not show an improvement in the mirror condition at present, need more practice before effects can be detected.

In conclusion, the current study showed that for children and adolescents with hemiplegia matching with the impaired hand is more accurate in a bimanual than in a unimanual matching condition. Similarly, mirror visual feedback had a positive effect on movement accuracy of the impaired arm, however, only in a subset of the individuals with SHCP. This variability in response may be related to differences in size and location of the brain lesions of the CP population and/or to the initial position sense of the impaired arm. Further research examining the relation between spasticity, position sense and improvements due to mirror visual feedback together with advanced brain imaging is warranted to determine which children might benefit most from bimanual practice with mirror visual feedback.

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Appendix A

The Tardieu Scale measures spasticity using two parameters: the spasticity angle and the spasticity grade (Gracies et al., 2010). The spasticity angle is the difference between the angles of arrest at slow speed and of catch-and-release at fast speed. The spasticity grade is an ordinal variable that grades the intensity and measures the muscle's reaction to fast passive stretch.

In this study we used the spasticity grade as an indication for the level of spasticity. Gracies et al. (2010) showed for this measure high intrarater and interrater reliability for experienced raters; 90% ± 8% and 81% ± 13%, respectively.

The Functional Independence Measure for children (WeeFIM) includes 18 items covering six areas in two dimensions (i.e., motor and cognitive). Motor: self-care (eating, grooming, bathing, dressing upper body, dressing lower body, toileting); sphincter control (bladder management, bowel management); transfer (chair/bed/wheelchair transfer, toilet transfer, tub/shower transfer); locomotion (crawling/walking/wheelchair, stair climbing). Cognitive: communication (comprehension, expression) and social cognition (social interaction, problem solving, memory; Sperle, Ottenbacher, Braun, Lane, & Nochajski, 1996; Tur et al., 2009). In the present study we only used the motor items of the WeeFIM. Ottenbacher et al. (1996) showed high test-retest responses for the WeeFIM with an intraclass correlation coefficient of 0.97.

The Manual Ability Classification System (MACS) is designed to classify how children with CP use their hands for object handling in daily life (Eliasson et al., 2006). It reports the collaboration of both hands together and is not an assessment of each hand separately. As shown in the study of Eliasson et al. (2006), the MACS has a good validity and reliability: intraclass correlation coefficient between therapists was 0.97.

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Deficits in upper limb position sense of children with Spastic Hemiparetic Cerebral Palsy are distance-dependent

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ABSTRACT

This study examined the arm position sense in children with Spastic Hemiparetic Cerebral Palsy (SHCP) and typically developing children (TD) by means of a contralateral matching task. This task required participants to match the position of one arm with the position of the other arm for different target distances and from different starting positions. Results showed that children with SHCP exhibited with both arms larger matching errors than the TD group, but only when the distance between the arms at the start of the movement was large. In addition, the difference in errors between the less-impaired and the impaired limb changed as a function of the distance in the SHCP group whereas no interlimb differences were found in the TD group. Finally, spasticity and restricted range of motion in children with SHCP were not related to the proportion of undershoot and size of absolute error. This suggests that SHCP could be associated with sensory problems in conjunction with their motor problems. In conclusion, the current study showed that accurate matching of the arms is greatly impaired in SHCP when compared to TD children, irrespective of which arm is used. Moreover, this deficit is particularly present for large movement amplitudes.

1. Introduction

Proprioception refers to the sense of body parts in space and comprises a static (sense of static limb position or position sense) and a dynamic component (sense of movement or kinaesthesia). It is a complex somatosensory modality that is imperative for the control of movement.

A large body of evidence details the critical role of proprioception in controlling muscle interaction torques (e.g. Sainburg, Ghilardi, Poizner, & Ghez, 1995) in timing the coordination between limb segments (Cordo, Carlton, Bevan, Carlton, & Kerr, 1994), in monitoring movement trajectories (Ghez, Gordon, Ghilardi, Christakos, & Cooper, 1990), and in establishing internal representations used during the acquisition and adaptation of skilled movement (Kawato & Wolpert, 1998). It is therefore not surprising that impaired proprioception is often suggested to be implicated in motor dysfunction such as in Parkinson's disease (Adamovich, Berkinblit, Hening, Sage, & Poizner, 2001), hemiparetic stroke (Niessen, Veeger, Koppe, Konijnenbelt, van Dieen, & Janssen, 2008), cerebellar disorders (Cody, Lovgreen, & Schady, 1993) or cerebral palsy (CP) (Cooper, Majnemer, Rosenblatt, & Birnbaum, 1995; Opila-Lehman, Short, & Trombly, 1985). Still, to facilitate the design of tailored therapeutic interventions, empirical research is required to get a detailed and more complete view of the deficits encountered by disabled individuals.

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A number of studies have already shed light on proprioception in CP. CP is a group of permanent disorders of movement and posture due to a non-progressive lesion in the fetal or infant brain (Miller, 2007). In children with Spastic Hemiparetic CP (SHCP) impaired control of muscle tone and spasticity in the limbs on one side of the body (the impaired side) severely complicates normal daily movement function. These deficits in daily functioning become predominantly evident for movements executed with the arm, which is usually more affected than the lower extremity (Charles & Gordon, 2006). Goble, Hurvitz, and Brown (2009) examined joint-position sense in this population using an arm flexion/extension task. This task required the participants to match the position of the elbow (occluded from view) to a target position to which elbow had been extended passively before the start of the trial. Larger errors were made with the impaired limb than with the less-impaired limb, and the latter was as accurate as the limbs of typically developing (TD) control children. It should be noted however, that in a sub-sample of the CP-population the condition is accompanied with memory deficits (Bottcher, 2010; Kolk & Talvik, 2000), which may have contributed to the reduced ability to match a previously felt position and complicates the interpretation of the results. Indeed, the contrasting findings of Chrysagis, Skordilis, Koutsouki, and Evans (2007) who showed with a similar task that children with SHCP made significantly larger errors than TD children with the impaired as well as the less-impaired arm, might be due to differences in the children's ability to memorize positions. Wingert, Burton, Sinclair, Brunstrom and Damiano (2009) used an alternative approach and tested joint-position sense using a forearm pronation/supination task in which the position of the occluded hand was to be aligned with a visual target. The 'cross-modal matching' required in this task, i.e., mapping between visual and proprioceptive information, adds another degree of difficulty (e.g. von Hofsten & Rosblad, 1988; Wann, 1991) and again implies that this task cannot be completed using somatosensory information only. In agreement with other work, this study showed that larger errors were made with the impaired limb than with the less-impaired limb. However, the overall performance of the hemiplegic group did not differ from the control group. Taken together, it thus seems that the accuracy of the joint-position sense (and the associated proprioceptive cues) is dependent on the joint (and the related muscle group) tested. In addition, these studies illustrate that it is difficult to assess joint-position sense in isolation (i.e. without confounding factors such as memory load or multi-modal mapping). Still, one aspect of joint-position sense that has not been considered in the study of SHCP is the ability to match the position of limbs in a contralateral matching task where the participant is instructed to copy the position of one limb by placing the other, contralateral limb, in the same mirror symmetric position. Such an intra-modal matching test, which does not require re-mapping between sensory inputs and in which the involvement of memory is considerably reduced, can provide us with useful information about how problems with proprioception influence tasks that involve both arms. This is particularly relevant for the study of children with SHCP whose motor impairments appear to be limited to one body side, but are known to hamper bimanual actions (Charles & Gordon, 2006). Therefore, in this study we will explore to what extent matching movements, in which both hands are involved, are hindered in children with SHCP by means of a contralateral matching task.

It has been suggested that position sense is dependent on the location (relative to the body) at which the measurement is performed. Localization of the hand is more precise in proximity of the body (i.e. at smaller distances relative to the body) than at larger distances from the body (van Beers, Sittig, & Denier van der Gon, 1998; Wilson, Wong, & Gribble, 2010). This phenomenon has been reported in studies of young (Goble & Brown, 2008; Goble, Lewis, & Brown, 2006) and elderly (Adamo, Martin, & Brown, 2007), supporting the notion that this effect is common and probably robust against neurodegeneration. van Beers et al. (1998) suggested that better localization at distances closer to the body may be understood from the geometry of the arm, alongside anatomical and physiological properties such as the fact that the number of muscle spindles acting about the joints in the arm increase in proximal direction (Scott & Loeb, 1994; c.i. van Beers et al., 1998). Verifying whether the accuracy in a proprioceptive-guided matching task in children with SHCP follows a similar trend (i.e. decrease in precision for locations further away from the body) may thus serve to test whether they are subject to similar anatomical and physiological constraints and use similar cues to localize the position of their hands as compared to TD children. To the best of our knowledge, this aspect has been largely overlooked in previous research into position sense of children with SHCP.

The aim of this study was therefore to add to the existing body of knowledge on proprioception in children with SHCP, and more specifically to gain insight into the accuracy of position sense of the impaired and less-impaired arm in a contralateral matching task. In a case study ($N = 2$) using a similar task Lee, Daniel, Turnbull, and Cook (1990) found that children with SHCP experienced difficulties with matching for both the impaired and less-impaired arm. The purpose of the current study was to substantiate these findings. In addition, considering the location-dependent effect on position sense, this study aimed to examine whether the accuracy of matching performance and possible differences between the SHCP and TD group on a contralateral matching task are location-dependent (i.e. dependent on the distance relative to the body). If the distance effect in children with SHCP does not significantly deviate from TD children, this could suggest that both groups use similar sensory cues to localize the hand and are subject to similar anatomical and physiological constraints, despite possible disturbances in the input and/or processing of sensory information.

2. Methods

2.1. Participants

Fourteen children with SHCP participated in this study (mean age 12.5 ± 1.9 years) of which six had a right and eight had a left hemiplegia (see Table 1 for further details). The participants were free from any neuromuscular disorders other than CP, did not have visual impairments or pain in either of the upper limbs, and they were not treated with Botulinum toxin in the past six

Table 1
Participant characteristics of the SHCP group. For each participant the age in years, sex, dominant hand, WeeFIM score, MACS level, Tardieu Scale, aetiology, and the maximum reaching distance (MRD) for the dominant and non-dominant arm are presented.

Participant	Age	Sex	Dominant hand[a]	WeeFIM/MACS	TS[b]	Aetiology	MRD D/ND[c]
1	13.4	M	Right	78/3	2	O_2 shortage during birth	41/27.2
2	10.5	M	Right	88/3	2	Cerebral infarction	47/30
3	10.8	M	Right	91/2	1.5	Unknown	33/31.5
4	14.5	M	Right	62/3	2	Schizencephaly	48/36.5
5	13.6	M	Right	91/2	2	Cerebral infarction	34/31.5
6	10.8	F	Right	52/3	1.5	Cerebral haemorrhage	31/26
7	12.1	F	Left	91/3	1	Cerebral infarction (thalamus)	46/42
8	15.5	M	Left	76/1	2	Unknown	47/46.5
9	9.3	M	Left	91/1	1	Cerebral infarction	25.5/24.5
10	13.1	F	Left	91/2	2	Cerebral infarction	39/38
11	14.4	M	Left	81/2	1	Cerebral haemorrhage	33.5/24.5
12	12.5	M	Left	59/3	2	Cerebral infarction	34/22.2
13	14.3	M	Left	71/3	2	Unknown	38/36.5
14	10.6	M	Left	87/2	0.5	O_2 shortage during birth	31/30.3

[a] The dominant hand is the less-impaired hand.
[b] Tardieu Score = mean of the individual scores of the biceps and the triceps.
[c] MRD, maximum reaching distance; D, dominant/less-impaired limb; ND, non-dominant/impaired limb.

months preceding the measurement. The children with SHCP were recruited through the Dutch society for children with a physical handicap and their parents. Before the actual start of the experiment, the Manual Ability Classification System (MACS), Functional Independence Measure (WeeFIM) and Tardieu score for spasticity were defined for the SHCP group in order to get an indication of the severity of the disorder (Table 1). The MACS describes how children use their hands during object handling and their need for assistance to perform manual skills in everyday life (Eliasson et al., 2006). The severity of performance limitation and the degree of required assistance increases for each MACS level from I to V. Seven children were classified in MACS level 3, five children in level 2 and two children in level 1. The WeeFIM scores range from 1 to 91 with a higher score representing a better functional independence. In the current population the WeeFIM scores ranged from 52 to 91. Finally, the Tardieu score was determined by a qualified physiotherapist as an indication of the children's spasticity. Individual scores were measured for the biceps brachii brevis and the triceps brachii longus and combined into one total score. All children showed mild to moderate spasticity with Tardieu scores ranging from 0.5 to 2.

In addition, a reference group of 20 TD children without any history of neuromuscular disorders and within the same age range as the children with SHCP (mean age 12.9 ± 2.6 years) were recruited among the Universities staff's families and friends. The TD children all had normal or corrected-to-normal vision and all but one were right hand dominant (determined by means of the Edinburgh Handedness Inventory (Oldfield, 1971)). Participant characteristics can be found in Table 1 (SHCP) and Table 2 (TD). Prior to testing the participant's parents provided written informed consent. All procedures were approved by the institutional research ethics committee and were in accordance with the Declaration of Helsinki.

Table 2
Participant characteristics of the TD group. For each subject the age in years, sex, dominant hand, score of the Edinburgh Handedness Inventory, and the maximum reaching distance (MRD) for the dominant and non-dominant arm are depicted.

Participant	Age (years)	Sex	Dominant hand	EHI score[a]	MRD D/ND[b]
1	13.0	M	Right	100	42/41
2	13.2	F	Right	100	37/37
3	12.3	F	Right	100	33/35
4	13.4	M	Right	100	36/34.5
5	8.3	F	Right	89	30/29
6	10.0	F	Right	80	30.5/29.5
7	16.9	F	Right	100	33.5/32.5
8	12.9	F	Right	90	34/33
9	13.3	F	Right	90	36/34
10	15.1	M	Right	90	40/40
11	11.4	M	Right	50	36/37
12	16.3	F	Right	40	32.5/34
13	10.9	F	Right	70	32.5/32.5
14	12.1	F	Right	60	38/37
15	16.5	F	Right	100	42/42
16	17.4	F	Right	70	35.5/34.5
17	14.9	M	Right	70	34/34
18	10.6	F	Right	100	28/27
19	10.6	M	Right	100	40/40
20	10.1	F	Left	−50	31/30

[a] EHI score, Edinburgh Handedness Score. +100 is complete right handedness; −100 is complete left handedness. If EHI was between −50 and +50 (ambidexter), the writing hand was identified as the dominant hand.
[b] MRD, maximum reaching distance; D, dominant limb; ND, non-dominant limb.

2.2. Materials and procedure

The child was seated on a height adjustable chair without armrests at a height adjustable table with the knees 90° flexed. Position sense was assessed using a custom made device consisting of two handles, each on a separate track fixed to a horizontal panel. The tracks were 20 cm apart, parallel to each other, and perpendicular to the medio-lateral axis of the trunk. The children were positioned such that the centre of the body was located in between the two tracks, and with the beginning of the track at 15 cm from the upper body. Vision of the limbs was blocked with an opaque cover on top of the wooden construction. The experimental setup is depicted in Fig. 1. The position of two parallel handles outside the box was recorded using one Optotrak unit with three infrared cameras (3020 Optotrak, Northern Digital Inc., Waterloo, Canada), which enabled us to calculate the position of the hands inside the box.

Before the start of the actual experiment, the maximum reaching distance of both arms was determined (MRD) in order to scale the different matching positions across subjects. MRD corresponds to the distance from the start of the track (position most proximal to the body) to the position of the handles when the elbows were extended as far as possible without bending the trunk forward. The MRD was used to determine the three target positions to be tested in the matching task, i.e., 25%, 50%, and 75% of the MRD. In case the MRDs of the left and right arm were different, the three target positions were based on the smallest MRD (this was applied for both groups). This means that for the children with SHCP the target positions were always based on the MRD of the impaired arm. The MRDs for each individual are reported in Tables 1 and 2.

The contralateral matching task required participants to match the position of one limb (reference limb), which was moved to the predetermined target position passively, by actively moving the other limb (matching limb) to the (mirror symmetric) position at the same distance as the reference arm. Three target positions (25%, 50%, and 75% of the MRD) were tested and the matching was done with either the less-impaired limb (dominant for TD children) or the impaired limb (non-dominant for TD children). The matching limb started at MRD (distally) or at the beginning of the track (proximally). The combination of all independent variables (3 target positions of the reference limb, 2 matching limbs, and 2 start positions of the matching limb), resulted in 12 trial types. Each trial type was performed once. The total amount of trials was divided in two blocks: (1) matching with the impaired (non-dominant) arm, and (2) matching with the less-impaired (dominant) arm. The order of blocks was randomized over participants and within each block the order of the trial types was randomized to reduce possible thixotropic effects on the matching accuracy (Proske, 2006). Prior to data collection 3 practice trials were conducted to familiarize the participant with the test setup and to check if the children were able to perform the movement properly. If the participant was unable to grip the handle due to his/her physical impairment, the experimenter placed the hand on top of the handle. However, in none of the participants the handle slipped out of the hands during a trial. In order to keep the children motivated they were told that the better their performance the more points they would earn. At the end of the experiment they could trade their points for a small gift.

2.3. Data analysis

The position data of the reference and the matching limb were imported into Matlab (version 7.1, The Mathworks Inc.). Then, absolute endpoint error was determined as the distance between the two handles at the end of the movement using custom-written routines. The end of the movement was verified by visual inspection of the plot showing the time series of the matching limb's position (inter-rater reliability $r = 0.98$, $p < 0.001$).

In addition, we calculated the proportion of trials in which the matching arm overshot or undershot the position of the reference target, resulting in amplitudes that were larger or smaller than the actual reaching distance respectively.

2.4. Statistical analysis

The MRDs of the SCHP group and the TD group were compared with a two-way repeated measures ANOVA with Limb (dominant/less-impaired, non-dominant/impaired) as a within factor and Group (SHCP, TD) as a between factor. The endpoint error in the contralateral matching task was analysed using a four-way repeated measures ANOVA with Limb (non-dominant/impaired, dominant/less-impaired), Position of the reference limb (25%, 50%, 75% MRD; i.e. the distance relative to the body), and Start position (distal, proximal) as within subjects factors and Group (SHCP, TD) as a between subjects factor. In case the sphericity assumption was violated, Greenhouse–Geisser adjustments were made. Fishers' LSD was used for post hoc analysis. To compare the proportions of undershoots and overshoots, a non-parametric Mann–Whitney *U* test was performed on the relative number of undershoots between the TD and the SHCP group. The significance level was set at 0.05.

3. Results

3.1. Maximum reaching distance (MRD)

A Limb × Group interaction ($F_{1,32} = 17.31$, $p < 0.001$) revealed that in children with SHCP the MRD of the less-impaired limb was larger than the MRD of the impaired limb ($p < 0.001$; 37.7 cm vs. 31.9 cm), while no such difference

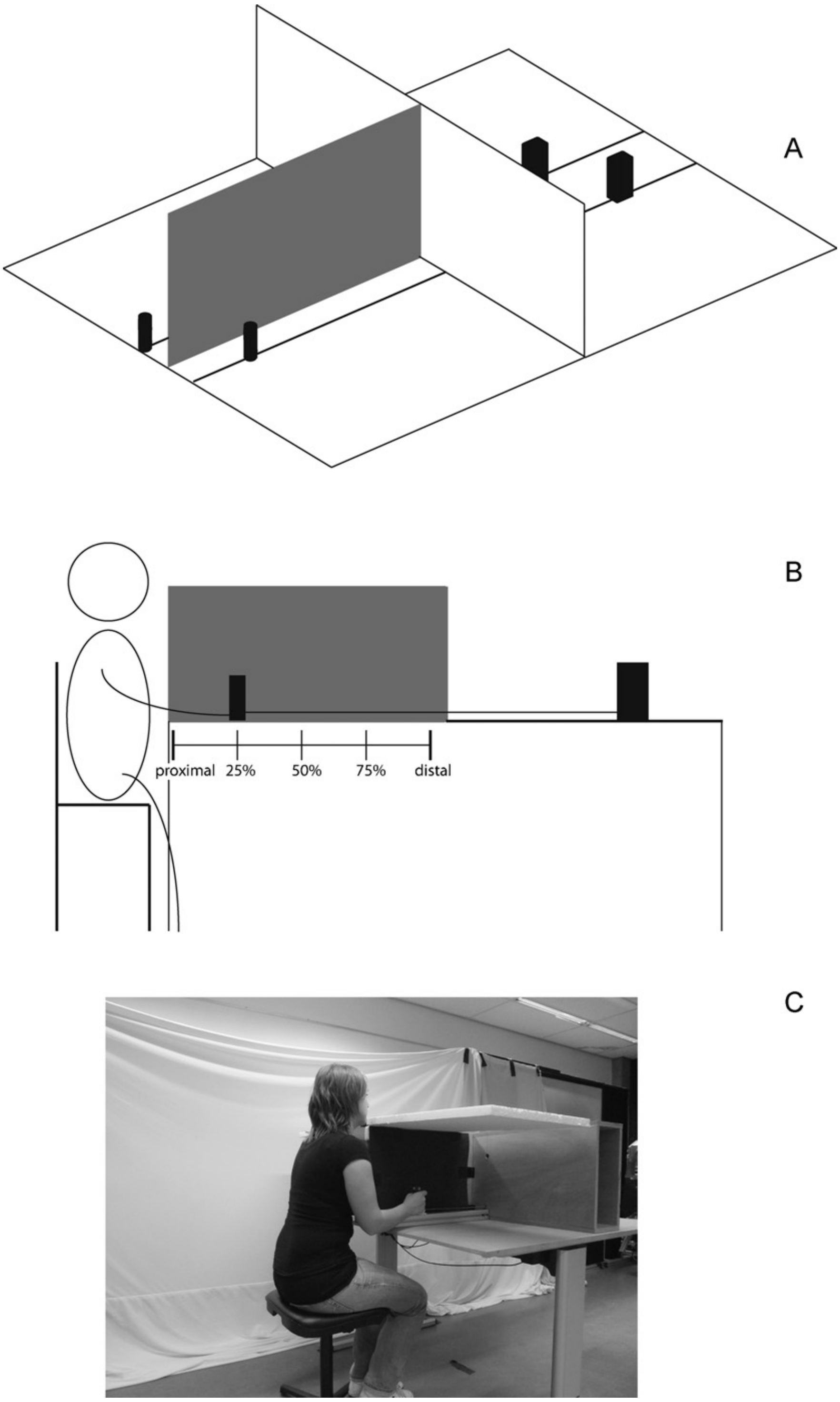

Fig. 1. (A) Top view of the experimental setup with the two handles that could be slid back and forth along the track. The screen between the arms prevented the hands from touching each other. The position of the handles outside the box was measured with an Optotrak camera (not depicted here). In this picture the opaque cover on top of the construction is not visible. (B) Side view of the experimental setup. The starting positions (proximal, distal) and the three target positions (25% MRD, 50% MRD, 75% MRD) are indicated. Please note that the target positions and the distal start positions (MRD) were determined based on the Maximum Reaching Distance of the child and thus differed per participant. (C) Real-life picture of the experimental setup with an opaque cover on top of the construction.

was found in TD children ($p = 0.63$; 35.1 cm vs. 34.7 cm, for dominant and non-dominant arm respectively). Further post hoc analysis of the Limb × Group interaction did not show differences in MRD between the limbs of the SHCP group and the limbs of the TD group (dominant arm: 37.7 cm (SHCP) vs. 35.1 cm (TD); non-dominant arm: 31.9 cm (SHCP) vs. 34.7 cm (TD)).

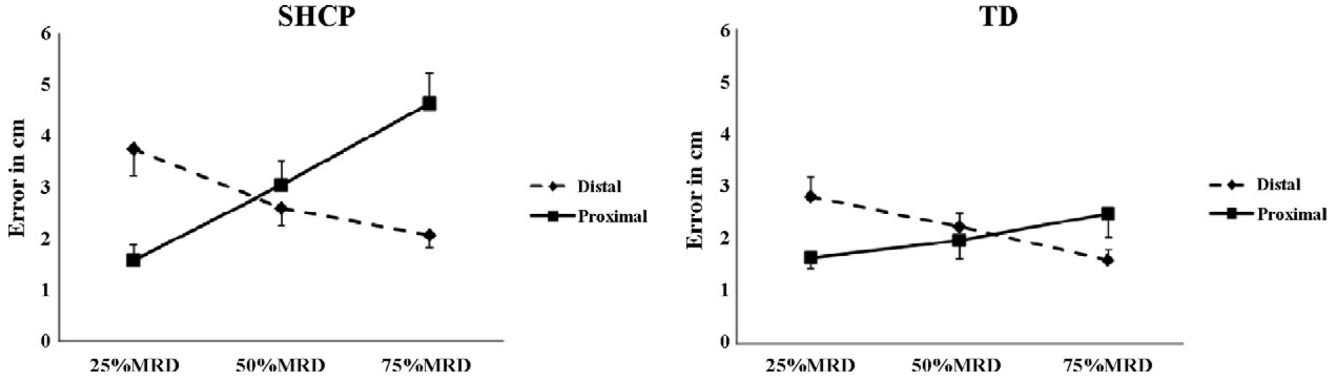

Fig. 2. The absolute endpoint errors (in cm) on different positions of the reference limb (25% MRD, 50% MRD, 75% MRD) for the different starting positions (distal, proximal) for the SHCP (left graph) and the TD group. The solid line represents the errors when the matching position was at 25%, 50% or 75% MRD when starting the movement proximally to the body. The dashed line represents the errors when matching the arms at a target position at 25%, 50% and 75% MRD when starting the movement distally from the body.

3.2. Endpoint error

All children were able to complete the experiment, but due to technical problems with the motion capture system during a number of trials of participants 7 (1 trial), 11 (2 trials), and 12 (2 trials) of the SHCP group, the data of these participants could not be included in the statistical analysis.

Analysis of the absolute error in the matching task revealed a two-way interaction between the factors Position reference and Start position ($F_{2,58} = 32.7$, $p < 0.001$), which was also present in two three-way interactions: Position reference × Start position × Group ($F_{2,58} = 5.3$, $p = 0.008$) and Position reference × Start position × Matching limb ($F_{2,58} = 3.4$, $p = 0.04$). Inspection of this Position reference × Start position interaction (see Fig. 2) showed an almost symmetrical picture for trials starting at a distal point and trials starting in proximity of the body, for both groups. Absolute error at 25% MRD in trials starting in the proximity of the body (i.e. 0% MRD) was similar to the absolute error at 75% MRD in trials starting at the most distal point from the body (100% MRD). Likewise, absolute error at 75% MRD in trials starting proximal to the body (i.e. 0% MRD) was not different from absolute error at 25% MRD in trials starting at the most distal point from the body (100% MRD). Finally, a distal or proximal start of the matching limb did not affect the amplitude of the error when the reference limb was positioned at 50% MRD. In fact, this Position reference × Start position interaction reveals a Distance effect indicating gradually larger absolute errors for larger reaching distances, i.e., the distance that has to be covered by the matching hand in order to achieve an error of 0. A secondary 3-way repeated measures ANOVA (Limb × Distance × Group), in which the dependent variables Position reference and Start position were combined into one factor (Distance), yielded identical results as the initial 4-way ANOVA (Fig. 3 explains the relation between the factors Position reference and Start position and Distance). For reasons of clarity and comprehensibility, the results of the secondary analysis, in which all participants were included, will be presented here.

This secondary analysis revealed main effects of Group ($F_{1,32} = 72.4$, $p = 0.002$) and Distance ($F_{2,64} = 29.5$, $p = 0.002$) on absolute error, which were superseded by a Group × Distance interaction ($F_{1.4,\ 44.3} = 5.5$, $p = 0.006$; see Fig. 4) and a Group × Distance × Limb interaction ($F_{2,64} = 3.8$, $p = 0.028$; see Fig. 5). Post hoc examination showed that the accuracy in this matching task dropped as a function of the reaching distance in both groups, but this drop in accuracy (i.e. increase in error) was significantly greater in the children with SHCP than in the TD children. This finding was further supported by the fact that there was no difference in absolute error between the SHCP and TD children for the small distance. In the medium distance the less-impaired limb of the SHCP group showed larger errors than the dominant arm of the TD group whereas no differences between the impaired arm and the non-dominant arm were found. Finally, when the reaching distance was large the errors made by both the impaired and the less-impaired arm were larger than in their counterparts of the TD group. Furthermore, no difference between the arms was found in TD children. In children with SHCP, however, matching with the impaired arm resulted in significantly larger absolute errors than matching with the less-impaired arm for the large distance condition (5.25 cm vs. 3.99 cm), while the opposite was found for the medium distance condition (2.64 cm vs. 3.93 cm). There was no difference between the impaired and less-impaired matching limb when the reaching distance was small.

3.3. Relative number of undershoot and overshoot

The proportion of trials resulting in an overshoot or undershoot is depicted in Table 3. All children undershot the target in the majority of the trials (TD: 80.8%, SHCP: 74.1%). These proportions were not significantly different ($U = 103.0$, $z = -1.31$, $p = 0.19$, average ranks = 19.4 and 14.9 for TD and SHCP respectively). In addition, inspection of Table 3 shows that the relative number of undershoots increased with increasing distance in both groups. The differences in the proportion of undershoots between the arms were small, especially in the SHCP group.

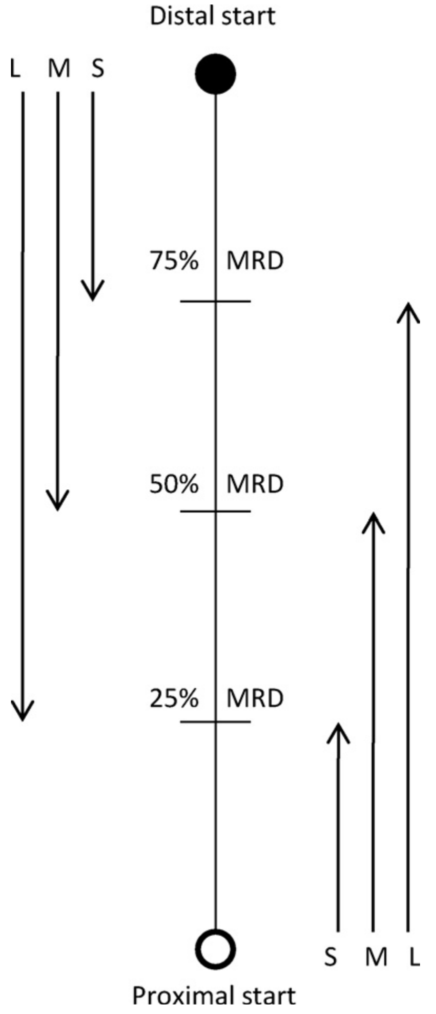

Fig. 3. Conversion from Position reference (25% MRD, 50% MRD, 75% MRD) and Start position (proximal, distal) into Distance (small (S), medium (M), and large (L)). It can be seen that e.g. moving towards 25% MRD when starting proximally results in the same distance as moving towards 75% MRD when starting distally.

3.3.1. Relation with the level of spasticity and MRD

Two additional analyses were performed in order to examine whether the level of spasticity (Tardieu score) and the difference in MRD between the limbs have an influence on the magnitude of the absolute errors and to the number of trials with undershoot in children with SHCP.

For the first additional analysis, the children with SHCP were divided into two groups based on their spasticity level as indicated by the Tardieu score. One group ('mild spasticity group') included all children with a Tardieu score equal to or below 1 ($n = 4$) and the other group ('moderate spasticity group') included the children with a score above 1 ($n = 10$). The results of the Mann–Whitney U test revealed that the 'mild spasticity group' did not differ significantly from the 'moderate spasticity group' on the percentage undershoots ($U = 15.5$, $z = -0.65$, $p = 0.51$, average ranks = 8.6 and 7.0 respectively). Likewise, no differences between the group with scores equal to or below 1 and the group with scores above 1 were found for the absolute error when matching with the impaired limb on all three distances (small: $U = 14.0$, $z = -0.85$, $p = 0.39$, average ranks = 6.0 vs. 8.1; medium: $U = 13.0$, $z = -0.99$, $p = 0.32$, average ranks = 9.3 vs. 6.8; large: $U = 10.0$, $z = -1.14$, $p = 0.16$, average ranks = 10.0 vs. 6.5).

For the second additional analysis, we compared the children with SHCP based on the relative difference of MRD between the less-impaired and the impaired arm. For each individual, the difference between the two MRDs (see Table 1) was divided by the largest MRD (expressed as a percentage) in order to minimize the inter-individual variability in arm length. The first group included the children with less than 10% relative difference ($n = 8$) and the second group included children with more than 10% relative difference ($n = 6$).

When comparing these two groups on relative number of undershoots, the Mann–Whitney U test did not reveal a significant difference between the groups ($U = 14.5$, $z = -1.26$, $p = 0.21$). The 'more than 10% group' showed an average rank of 5.9 and the 'less than 10% group' had an average rank of 8.7. This then suggests that both groups did not significantly differ on relative numbers of undershoot.

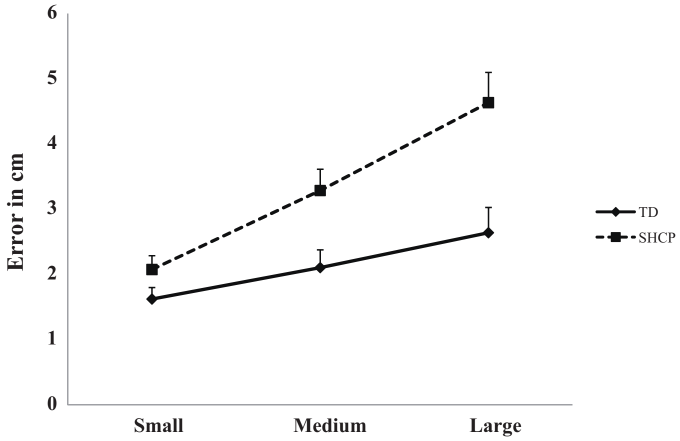

Fig. 4. The absolute errors (in cm) of the typically developing (TD) and the cerebral palsy (SHCP) group for the different distances (small, medium, and large). The dashed line represents the errors of the SHCP group and the solid line represents the errors of the TD group.

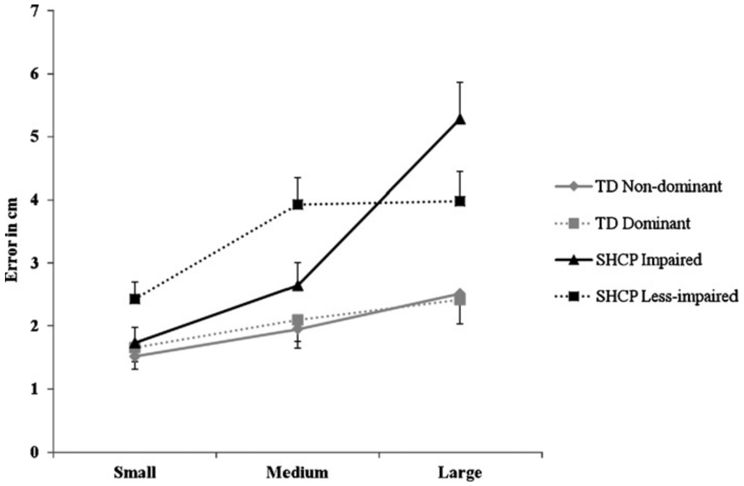

Fig. 5. The absolute errors (in cm) of both upper limbs for the Typically Developing (TD) group (grey lines) and the Spastic Hemiparetic Cerebral Palsy (SHCP) group (black lines) depticted for each distance separately. The distances (small, medium, large) are depicted on the horizontal axis. The solid grey line represents the errors of the non-dominant arm, the dashed grey line represents the errors of the dominant arm. The error of the impaired arm of the SHCP group are depicted with a solid black line and the error of the less-impaired arm is represented by the dashed black line.

Table 3
Percentages (and proportions) of the overshoots and undershoots in the SHCP (impaired and less-impaired arm) and the TD group (non-dominant and dominant arm) in the small, medium and large distance. In the last column the total relative number of under- and overshoots is depicted. The range (in cm) of the total percentage overshoots (positive values) and undershoots (negative values) is indicated between brackets.

	Small	Medium	Large	Total
Undershoot				
SHCP				
Impaired	66.7% (18/27[a])	63% (17/27)	89.3% (25/28)	74.1% (−18.3 to −0.1)
Less-impaired	73.1% (19/26)	71.5% (20/28)	81.5% (22/27)	
TD				
Non-dominant	67.5% (27/40)	70% (28/40)	87.5% (35/40)	80.8% (−7.0 to −0.01)
Dominant	80% (32/40)	87.5% (35/40)	90% (36/40)	
Overshoot				
SHCP				
Impaired	33.3% (9/27[b])	37% (10/27)	10.7% (3/28)	25.9% (0.03 to 5.7)
Less-impaired	26.9% (7/26)	28.5% (8/28)	18.5% (5/27)	
TD				
Non-dominant	32.5% (13/40)	30% (12/40)	12.5% (5/40)	19.2% (0.02 to 3.3)
Dominant	20% (8/40)	12.5% (5/40)	10% (4/40)	

[a] Number of trials with undershoot/total number of trials.
[b] Number of trials with overshoot/total number of trials.

Also when focusing on the absolute error, no differences were demonstrated. The absolute error on the small distance when matching with the impaired limb showed similar ranks for the groups with large and the small differences in MRD ($U = 22.0$, $z = -0.26$, $p = 0.8$, average ranks 7.2 and 7.8 respectively). Also for the medium and the large distance no differences were found between the 'less than 10% group' and the 'more than 10% group' (medium: $U = 12.0$, $z = -1.55$, $p = 0.12$, average ranks = 5.5 vs. 9.0; large: $U = 15.0$, $z = -1.16$, $p = 0.25$, average ranks = 9.0 vs. 6.4).

4. Discussion

In order to better understand the impact of Spastic Hemiparetic Cerebral Palsy (SHCP) on position sense during bimanual tasks, the current study compared the performance of children with SHCP and TD children in a typical contralateral arm matching task. We found that children with SHCP matched the position of the reference arm less accurately than TD children as reflected in larger matching errors for both the impaired and less-impaired arm. Previously, Wann (1991) has shown similar bilateral deficits in a small group of children with mixed CP-diagnosis, i.e., quadriplegia and diplegia where the condition is caused by a lesion to the left and right hemisphere. Yet, our results demonstrate that also children with unilateral brain damage have difficulties with matching the position of the upper limbs (without visual information), which is in congruence with Lee et al. (1990) who reported similar findings in a case study with two children with SHCP. Interestingly, the performance in the current contralateral matching task appeared to depend on the range of the reaching movement required to match the target. In both the SHCP and the TD children endpoint error gradually increased as a function of the initial distance between the reference limb and the matching limb, i.e., at the start of the trial. In contrast to previous research that showed a drop in precision when localizing targets further away from the body (i.e., larger distance relative to the body) (e.g. Adamo et al., 2007; van Beers et al., 1998), the distance effect found in the current study was independent of the target position. Rather, the accuracy in this matching task was affected by the distance of the reaching movement irrespective of whether the movement was to a proximal or a distal target. It should be noted that this effect was stronger for the SHCP than for the TD children. In addition, further analysis showed that performance of the two groups only differed significantly in the medium and large distance condition.

What makes matching more prone to error when the initial distance between the effector and the target is larger? The cause of this distance effect might be related to the nature of movements children perform and practice as part of their daily routine. Daily movements in which both limbs are involved are usually movements in which the limbs are relatively close to each other, for example cutting a piece of bread, typing on the computer, or playing with a doll. As a result it is conceivable that the joint-position sense is better developed within the daily range of motion and less developed (less specific) outside that range. Furthermore, larger reaching movements are also more prone to signal-dependent noise as they require neural command signals of a greater intensity, which come with increased variance of noise (Goble, 2010; Harris & Wolpert, 1998). This phenomenon is expected to amplify the endpoint error of movements with larger amplitudes. In addition, for children with SHCP, involuntary muscle contractions associated with spasticity can lead to a situation in which the muscle tends to remain in a shortened position. This restriction in range of motion may cause length-related changes in the muscle-tendon complex and can eventually lead to a loss of joint range, or contracture (Ada, O'Dwyer, & O'Neill, 2006). Although spasticity may impede the movement required in the present study, it has to be noted that the movement was self-paced and within the range of motion of the impaired limb which should have limited the impact of the (high) velocity depended reaction. If the restriction in range of motion would explain the difference in matching accuracy between the SHCP and the TD group, more undershoot would be expected in the SCHP group (in particular for the spastic impaired arm) compared to the TD group. Yet, both groups undershot the target in the majority of the trials and there was no difference between the children with SHCP and the controls, or between the impaired and the less-impaired arm. Moreover, children with low levels of spasticity undershot the target in as many trials as the children exhibiting higher levels of spasticity and the size of the absolute error neither differed between these groups. A similar finding was demonstrated for the difference in MRD: the group with larger differences in MRD between the impaired and less-impaired arm did not show significantly more frequent undershoots or larger absolute errors than the group with smaller differences in MRD. Therefore, although we cannot exclude that the restricted range of motion in children with SHCP may have contributed to the larger endpoint errors at the large distance, the present results suggest that a compromised motor system cannot fully account for the lower matching accuracy in the SHCP group and the high prevalence of undershoot.

In addition to the diminished matching ability of the impaired arm, larger endpoint errors for the less-impaired arm compared to the dominant arm in the medium and large distance condition indicate, in agreement with previous research (Chrysagis et al., 2007; Goble, Hurvitz, et al., 2009; Wingert et al., 2009), that SHCP could be associated with sensory problems in conjunction with their motor problems. The performance in the contralateral matching task is the combined result of a number of interacting factors. Afferent proprioceptive signals determine the position of the reference arm. This information is processed at cortical level leading to efferent motor commands which move the contralateral arm to the felt target position. Finally, afferent proprioceptive signals coming from the matching arm may be used to fine tune and match the position of the reference arm. It is impossible to pinpoint the origin of a matching problem on the basis of our findings, however a detailed comparison of the performance of the impaired and less-impaired limb may provide more insight into the specific difficulties encountered by children with SHCP in tasks requiring bimanual control. A first question that needs to be addressed is whether the matching difficulties may be explained by a deficit at the cortical level only. A deficiency in mapping proprioceptive signals from the reference arm onto an egocentric reference frame is likely to result in distance independent matching errors for both arms, i.e., the matching error would be the same for both arms on each distance. However, the finding that performance of the limbs of children with SHCP was only comparable (with each other and with the TD group) in the small distance condition appears to be inconsistent with this notion and suggests that deficits occur both at cortical level and at the level of the muscle. Secondly, while the impaired arm located the target less accurately than the less-impaired in the large distance condition, the opposite was found in the medium distance condition. This is in contrast with the TD children where the endpoint error was similar for both arms in all three conditions and raises the question whether position sense may be affected in the less-impaired arm of children with SHCP too. Based on purely unimanual pointing tasks, Goble, Hurvitz, et al. (2009) and Wingert et al. (2009) concluded that position sense of the less-impaired arm was not reduced. The implication would then be that the larger matching errors of the less-impaired limb for the medium distance condition in our study were caused by disturbed afferent information originating from the impaired reference limb only. This would suggest that SHCP would affect the accuracy of position sense when the impaired limb is used as a static reference (or target) more than when it is actively involved in the reaching movement. However, given the fact that involuntary spastic contractions primarily emerge when the affected muscle is stretched (i.e. dynamic rather than static conditions) the aforementioned suggestion seems to be counterintuitive. Thus while decreased position sense of the impaired limb is likely to contribute to the matching errors of the less-impaired limb, at this moment the contralateral matching task does not allow us to exclude difficulties at the level of the less-impaired arm either. At last it should also be noted that in the current study the differences between the impaired and the less-impaired side may also be related to the fact that the target locations were based on the smaller maximum reaching distance of the impaired limb. This meant that the less-impaired limb operated within smaller range of movement relative to its maximal range than the impaired limb, which may be partly responsible for the smaller error of the less-impaired limb at large distances.

To summarize, although the contralateral matching task is unable to isolate position sense deficits of the impaired and less-impaired arm, the current results demonstrate that children with SHCP are clearly disadvantaged when performing skills that involve both arms. Accurate positioning of one arm relative to the position of the other arm, which is required in numerous manual skills, is impaired regardless of which arm is used.

Finally, it has been suggested that tasks requiring processing and mapping of proprioceptive information are subserved by a fronto-parietal network that is mainly located within the right hemisphere (Goble & Brown, 2008). This is consistent with findings of Goble, Hurvitz, et al. (2009) demonstrating poorer proprioceptively guided matching in individuals with right hemispheric damage than in individuals with a left hemispheric damage. Reinspection of our data (6 children with right hemispheric damage vs. 8 children with left hemispheric damage) did not reveal such a difference. Since we were unable to match these two groups for size and specific location of the lesion, caution is warranted when interpreting these results. Moreover, other findings of Goble show that left-handed individuals have a right hand advantage for proprioceptive tasks (Goble, Noble, & Brown, 2009), indicating that other factors related to practice and specific function of the hand are likely to contribute to the left–right differences in position sense. Altogether without controlling for important confounding factors, such as specific location of the lesion, size of the lesion, functionality of the impaired arm, etc., we believe it is premature to compare SHCP children with left and right hemispheric damage.

In conclusion, the results of the present study demonstrate that children with SCHP exhibit severe deficiencies in accurate positioning of one arm relative to the position of the other arm when compared to TD children. Despite the fact that with a contralateral matching task we cannot draw conclusions on the origin of the proprioceptive deficits, it is suggested that the unilateral proprioceptive deficits reported by previous studies, severely hamper the matching of the limbs. This deficit is particularly visible when the initial distance between the target and the matching arm is large (irrespective of target position relative to the body).

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The positive effect of mirror visual feedback on arm control in children with Spastic Hemiparetic Cerebral Palsy is dependent on which arm is viewed

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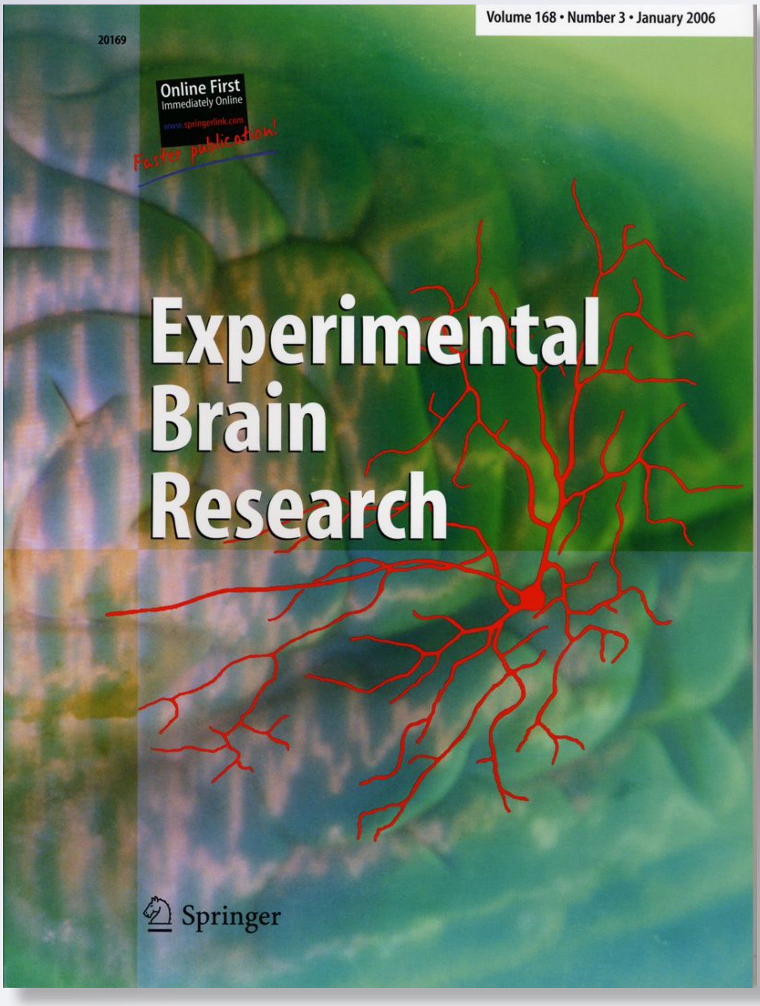

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RESEARCH ARTICLE

The positive effect of mirror visual feedback on arm control in children with Spastic Hemiparetic Cerebral Palsy is dependent on which arm is viewed

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Abstract Mirror visual feedback has previously been found to reduce disproportionate interlimb variability and neuromuscular activity in the arm muscles in children with Spastic Hemiparetic Cerebral Palsy (SHCP). The aim of the current study was to determine whether these positive effects are generated by the mirror per se (i.e. the illusory perception of two symmetrically moving limbs, irrespective of which arm generates the mirror visual feedback) or by the visual illusion that the impaired arm has been substituted and appears to move with less jerk and in synchrony with the less-impaired arm (i.e. by mirror visual feedback of the less-impaired arm only). Therefore, we compared the effect of mirror visual feedback from the impaired and the less-impaired upper limb on the bimanual coupling and neuromuscular activity during a bimanual coordination task. Children with SHCP were asked to perform a bimanual symmetrical circular movement in three different visual feedback conditions (i.e. viewing the two arms, viewing only one arm, and viewing one arm and its mirror image), combined with two head orientation conditions (i.e. looking from the impaired and looking from the less-impaired body side). It was found that mirror visual feedback resulted in a reduction in the eccentric activity of the Biceps Brachii Brevis in the impaired limb compared to the condition with actual visual feedback from the two arms. More specifically, this effect was exclusive to mirror visual feedback from the less-impaired arm and absent when mirror visual feedback from the impaired arm was provided. Across conditions, the less-impaired arm was the leading limb, and the nature of this coupling was independent from visual condition or head orientation. Also, mirror visual feedback did not affect the intensity of the mean neuromuscular activity or the muscle activity of the Triceps Brachii Longus. It was concluded that the positive effects of mirror visual feedback in children with SHCP are not just the result of the perception of two symmetrically moving limbs. Instead, in order to induce a decrease in eccentric neuromuscular activity in the impaired limb, mirror visual feedback from the 'unaffected' less-impaired limb is required.

Keywords Cerebral palsy · Hemiparesis · Mirror visual feedback · Neuromuscular activity · Electromyography · Bimanual coordination

Introduction

Children with Spastic Hemiparetic Cerebral Palsy (SHCP), who have unilateral motor impairments in both their arm

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and leg due to brain and/or pyramidal tract damage (Miller 2007),[1] perform tasks requiring only the less-impaired hand reasonably well (e.g. Steenbergen et al. 1996; Utley and Sugden 1998). In contrast, tasks requiring bimanual coordination pose a huge challenge because of the inevitable involvement of the impaired arm and hand. In recent years, bimanual reaching and grasping has been thoroughly investigated in individuals with SHCP (e.g. Utley and Sugden 1998; Volman et al. 2002; Sugden and Utley 1995; Steenbergen et al. 1996). Interestingly, these studies suggest that, despite the unilateral impairment, bimanual actions of children with SHCP seem to be facilitated by bilateral connections at multiple levels of the central nervous system similar to what has been found in typical populations (e.g. corticospinal, cerebellar, brain stem, and propriospinal; Wiesendanger et al. 1994). For example, Volman et al. (2002) showed that when drawing circles in an in-phase (symmetrical) coordination mode, the spatio-temporal interlimb variability decreased. Furthermore, movement smoothness of the impaired limb increased compared with single-handed performance. Steenbergen et al. (2008) observed close temporal synchrony of the hands when grasping an object bimanually, which contrasted with the timing differences between both hands when they performed separately. It should be noted that some of these findings indicate adaptations of the less-impaired side to the behaviour of the affected side (e.g. Steenbergen et al. 1996), but combined these studies suggest that bilateral interactions exist in children with SHCP and that they can lead to favourable effects in the impaired arm.

A paradigm that has been used to further our understanding of how visual and spatial processes influence coordination and perception of the two hands is the ‘mirror box illusion’ (e.g. Franz and Packman 2004; Holmes and Spence 2005). This illusion is manifested when a mirror is placed in between the two upper limbs along the midsagittal plane. The reflection of the arm viewed in the mirror seems superimposed on the visual image of the arm behind the mirror. When the arm facing the reflective side is moved, this creates the illusory perception of a zero lag symmetrical movement of the two limbs. The effects of mirror visual feedback were first investigated by Ramachandran and Rogers-Ramachandran (1996) in amputees with phantom pain. After a short period of ‘mirror box’ therapy, which involved (bilateral) mirror-symmetric movements, amputees reported a decrease in phantom pain. These encouraging findings led to the adoption of mirror visual feedback in treating other acquired unilateral motor or pain disorders where the illusion appeared to result in positive effects on motor performance and pain perception (for a review see Ramachandran and Altschuler 2009). For instance, it was found that chronic stroke patients could benefit from therapy using mirror visual feedback, showing increases in the range of motion, speed and accuracy of arm movements (Altschuler et al. 1999; Stevens and Stoykov 2003), an improved functional use and a recovery of grip strength (Sathian et al. 2000). Likewise, in patients with Chronic Regional Pain Syndrome 1 (CRPS1) mirror visual feedback of the unaffected limb reduced the perception of pain and stiffness (McCabe et al. 2003).

Interestingly, Feltham et al. (2010a, c) demonstrated that the positive effects of mirror visual feedback may potentially be extended to individuals with congenital disorders such as SHCP, a finding that was recently supported by Gygax et al. (in press) who showed that mirror therapy in children with hemiplegia may improve strength and dynamic function of the impaired arm. Feltham et al. (2010a, c) used a task where participants performed continuous symmetrical circular movements with both upper limbs in three visual conditions (glass: seeing the two arms; screen: seeing only the less-impaired arm; mirror: seeing the less-impaired arm and its mirror reflection). An effect of mirror visual feedback was found on the nature of the bimanual coordination (Feltham et al. 2010a) and on the neuromuscular activation in children with SHCP (Feltham et al. 2010c). More specifically, in the first study, it was demonstrated that movement variability of the interlimb coupling was lower in the mirror condition in comparison with the screen condition. In addition, mirror visual feedback resulted in a reduction in the neuromuscular intensity in the shoulder muscles of the less-impaired limb and a shortening of the duration of eccentric and concentric activity in the elbow muscles of the impaired limb. In accordance with Perry et al. (2001), a phase where a flexor muscle (e.g. Biceps Brachii Brevis, BBB) was actively contributing to a flexion movement was defined as *concentric*, whereas flexor activity was *eccentric* when it contributed to an extension movement. For extensor muscles (e.g. Triceps Brachii Longus, TBL), the opposite classification was used. Note that an earlier study showed that children with SCHP performed this bimanual coordination task with higher levels of neuromuscular intensity in elbow and wrist muscles and longer periods of concentric and eccentric activity in elbow and shoulder muscles compared with typically developing children (Feltham et al. 2010b). More eccentric activity of the BBB might suggest more counteraction to the extension movement and hence indicates that the neuromuscular control is less

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[1] Cerebral Palsy (CP) is a group of permanent disorders of movement and posture due to a non-progressive lesion in the foetal or infant brain (Miller 2007). CP is the most common cause of childhood disability and has an incidence of 2–2.5 per 1,000 living births (Lin 2003). A common form of CP is Spastic Hemiparetic Cerebral Palsy (SHCP). Children with SHCP have a brain lesion in one hemisphere and as a result have spasticity on the other side of the body.

efficient in children with SHCP. The finding of a decrease in interlimb variability and a reduction in eccentric and concentric muscle activity in a condition with mirror visual feedback thus shows that the mirror has the capacity to induce a general improvement of the kinematics and the neuromuscular efficiency during bimanual movements in children with SHCP.

A pertinent question is, however, whether the mirror effects observed in these children are caused by the illusory perception of seeing two arms moving in perfect symmetry, irrespective of which arm is seen in the mirror, or by the illusion that the impaired limb has been substituted with a less-impaired limb, which is not spastic. The studies by Feltham et al. (2010a, c) described above have only investigated the effect of mirror visual feedback from the unaffected arm and therefore were not able to discriminate between these two explanations. When Franz and Packman (2004) found that mirror visual feedback was powerful enough to enhance spatial coupling of the two hands in healthy adults performing a circle drawing task in a similar manner as actual vision of both hands, this effect was independent of the laterality of the mirror visual feedback. In a condition where only one hand was visible, the circles drawn by the hand in vision were found to be significantly larger than for the hand hidden behind the screen. Mirror visual feedback, regardless of which hand was viewed, had the capacity to wipe out this between-hand difference in circle size. Franz and Packman (2004) hypothesised that the illusion of the perfect symmetry between the two hands created by the mirror promoted the sensorimotor coupling at the central level.

In children with SHCP, however, the movement produced by the impaired and less-impaired arm is qualitatively different, and hence, the mirror visual feedback created by either arm is considerably different as well. Whilst there is an illusion of perfect symmetric movement in both situations, the mirror visual feedback of the impaired arm shows a less smooth movement hampered by the motor deficits. This discrepancy between the two sides and the mirror visual feedback they elicit enables us to investigate the mirror box illusion in this group of children in more detail. More specifically, the aim of the present study was to determine whether the mirror effects as found previously by Feltham et al. (2010a, c) are the result of the perception of visual symmetry per se, irrespective of which arm is viewed, or by the illusion that the impaired arm has been substituted and appears to move smoother and in synchrony with the less-impaired arm. For this purpose, we compared the effect of mirror visual feedback generated by the less-impaired and the impaired arm on the bimanual coupling and the neuromuscular activity in children with SHCP during a bimanual coordination task similar to the one used in Feltham et al. (2010a, c). Based on the studies of Feltham et al. (2010a, c) we anticipate that mirror visual feedback from the less-impaired arm will result in smaller interlimb variability and reduced eccentric activity in the arm muscles of the impaired limb compared to the visual feedback of both arms (glass condition). If the illusion of visual symmetry is the main trigger for the changes induced by the mirror, mirror visual feedback of the less-impaired arm is expected to induce similar effects on the kinematics and the neuromuscular activity as compared to mirror visual feedback of the impaired arm. Alternatively, if the mirror effect in children with SHCP is caused by a mechanism involving substitution of the visual information of the impaired arm by visual feedback from the less-impaired arm, we expect to find less favourable changes to the control of the movement when viewing the impaired upper limb and its mirror reflection than when viewing mirror visual feedback of the less-impaired limb.

Methods

Participants

Ten children (eight males and two females) with SHCP participated in the study (mean age 12.7 ± 3.2 years). Further participant characteristics can be found in Table 1. A subset of the data from seven children who took part in a previous study (Feltham et al. 2010c) was identified to be included in the present analysis. The participants did not have impaired vision or any neuromuscular disorders other than SHCP. Written informed consent was obtained from all participating children and their parents. The experiment was conducted in accordance with the Declaration of Helsinki, and all experimental procedures were approved by the institutional research ethics committee.

Test procedures

Each participant was seated on a height adjustable chair at a table with both feet flat on the floor and the knees 90° flexed. The elbows were flexed over 90°, and in each hand, the participant grasped a handle attached to a wooden disc (radius 0.10 m) which spun freely 360° around a vertical axis. The axes were fixed to a wooden plateau and were located 0.31 m apart.

Participants were asked to perform a continuous inward symmetrical circular bimanual movement (the right arm rotated anti-clockwise and the left arm rotated clockwise). Starting at the inner most part of each circle (9 o'clock for the right arm and 3 o'clock for the left arm), children were asked to rotate the discs continuously at a self-selected speed until they were instructed to stop. Additionally, they were instructed to keep the movement time per cycle (i.e.

Table 1 Participant characteristics

Participant	Age	Sex	Hand dominance	MAS	GMFCS	WeeFIM	Aethiology
1	12.8	M	Left	1	I	90	Unknown
2	9.3	F	Left	1+	I	89	Cerebral haemorrhage
3	13.2	M	Left	1	I	91	Unknown
4	14.3	M	Left	1+	I	91	Cerebral haemorrhage during birth and meningitis just after birth
5	11.0	M	Left	1	II	55	Meningitis just after birth
6	6.8	M	Left	1	I	83	O_2 shortage during birth
7	17.1	M	Left	2	I	91	Cerebral haemorrhage
8	11.1	M	Right	1	I	91	Unknown
9	14.7	M	Right	2	II	62	Schizencephaly
10	16.3	F	Right	1	I	79	O_2 shortage during birth

Severity of the impairment was assessed by a single experimenter with the Modified Ashworth Scale (MAS; spasticity levels increase from 1 to 4), Gross Motor Function Classification System (GMFCS; function deteriorates from I to V) and the functional independence measure for children (WeeFIM; motor items only, with a possible score range of 13–91. A higher score denotes a better functional independence of the child)

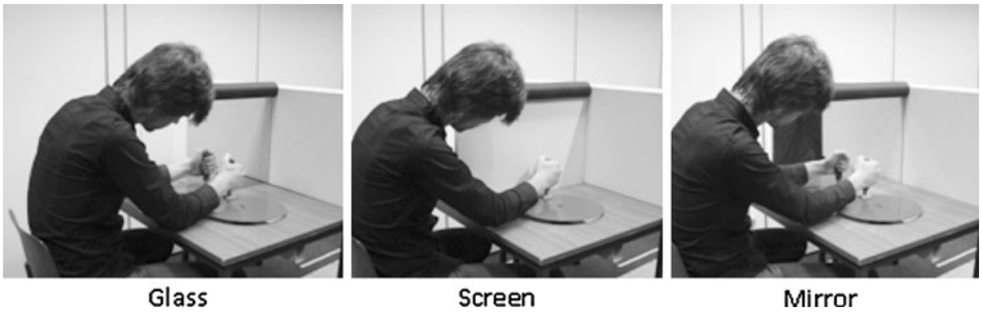
movement frequency) constant across the experimental trials and the different conditions.

The type of visual feedback was varied so that the participant (1) viewed both arms, (2) viewed only one arm and (3) viewed one arm and its mirror reflection, by placing a glass, opaque screen or mirror divide, respectively (all: width 0.06 m, depth 0.75 m, height 0.39 m), between the arms along the midsagittal plane (Fig. 1). The glass and the screen conditions were added as control conditions. In addition, in order to examine the difference between mirror visual feedback of the less-impaired arm (referred to as ‘uncompromised’ mirror visual feedback) and mirror visual feedback of the impaired arm (referred to as ‘compromised’ mirror visual feedback) on the nature of the bimanual coupling and the neuromuscular activity in the BBB and TBL muscle, the orientation of the head (i.e. viewing side) was varied; the participants orientated their head either towards the impaired side of the body (ViewImp) or to the less-impaired side of the body (ViewLessImp).

The six conditions (3 visual feedback × 2 viewing side conditions) were presented in a random order and per condition, three trials, each lasting approximately 15 s, were recorded. Prior to data collection, practice trials were conducted to familiarise the participants with the test setup. Short breaks were given between the trials in order to recover from any fatigue or decrease in concentration that might have occurred during the performance of the experiment. In order to keep the participants motivated, they were told that rotating the discs more symmetrically resulted in more points. At the end of the experiment, the children could trade their points for a small gift.

Recording and analysis procedures

The 3D position of the wrist, elbow and shoulder was determined by two serially connected units containing three infrared cameras at 200 Hz (3020 Optotrak, Northern Digital Inc., Waterloo, Canada). Light emitting diodes were bilaterally attached to the skin with double-sided tape over the dorsal



Glass Screen Mirror

Fig. 1 Experimental setup showing one of the experimenters demonstrating the task during the glass (*left panel*), screen (*middle panel*) and mirror (*right panel*) condition. The participant viewed the bimanual task either from the impaired or from the less-impaired side of the body. Note that the participants were considerably smaller than the experimenter and that their posture was more erect than shown in this picture tuberculum of the radius (wrist), lateral epicondyle of the humerus (elbow), greater tubercle of the humerus (shoulder) and the trochantor of the femur (hip). The phase of each limb was calculated according to the following formulas:

$$\varphi \mathrm{D} = \arctan\left[\left(\mathrm{d}S_{\mathrm{D}} \cdot \mathrm{d}t^{-1}\right)/S_{\mathrm{D}}\right],$$

and

$$\varphi \mathrm{ND} = \arctan\left[\left(\mathrm{d}S_{\mathrm{ND}} \cdot \mathrm{d}t^{-1}\right)/S_{\mathrm{ND}}\right],$$

where φD and φND are the phase of the dominant (less-impaired) and the non-dominant (impaired) hand, respectively, S_{D} and S_{ND} are the position time series, and $\mathrm{d}S_{\mathrm{D}}\cdot \mathrm{d}t^{-1}$ and $\mathrm{d}S_{\mathrm{ND}}\cdot \mathrm{d}t^{-1}$ represent the instantaneous velocity. Before the calculation of φND, the sign of the position time series of the non-dominant arm was inversed to an anti-clockwise trajectory. The continuous relative phase (CRP) indicating the degree of coupling (i.e. synchronicity) between the arms is then:

$$\mathrm{CRP} = \varphi \mathrm{D} - \varphi \mathrm{ND},$$

where a positive value for CRP implied the less-impaired arm lead and a negative value the impaired arm lead.

Superficial EMG (electromyography) was bilaterally recorded from the main muscles around the elbow: the Biceps Brachii Brevis (BBB) and the Triceps Brachii Longus (TBL), according to the SENIAM guidelines for surface EMG measurement (Hermens et al. 2000). The ground electrode was placed over the acromion on the side of the less-impaired hand. Disposable Ag/AgCl surface EMG electrodes with a gel-skin contact, active detection area of 15 mm^2 for each electrode and a 20 mm centre to centre inter-electrode distance, were placed in parallel with the muscle fibre direction over the muscle bellies after cleaning and gentle abrasion of the skin. The EMG signals were amplified 20 times, high-pass pre-filtered at 10 Hz and AD-converted at 1,000 Hz with a 22-bit resolution and stored on a computer. The EMG signals were band-pass filtered with a zero lag 2nd order Butterworth filter between 10 and 400 Hz and then full-wave rectified. Finally, the EMG signals were smoothed with a zero lag 2nd order low-pass Butterworth filter at 6 Hz.

Bilateral EMG recordings were analyzed from the first two cycles of each trial.[2] Typically, EMG amplitudes are scaled to the activation levels recorded either during an isometric maximal voluntary contraction or a specified steady-state sub-maximal contraction. However, this procedure is likely to be unreliable in people with neurological conditions since they are often unable or unwilling to perform maximum contractions (van Dieën et al. 2003; Smith et al. 2008). Therefore, to determine the intensity of the mean neuromuscular activity of each muscle during the bimanual movement, the mean amplitude was calculated from the smoothed raw EMG signals. In addition, the amount of concentric and eccentric muscle activity was determined. To this end, the EMG profile of each muscle was broken down into active and inactive phases, after the threshold for muscle contraction was determined. Consistent with Perry et al. (2001), it was assumed that a purposeful activation of a muscle causes an increase in the EMG signal within the frequency range of 0–160 Hz. The active/inactive threshold value was then calculated as follows: $T = 15 + 1.5R$, where T is the threshold value, R is the mean value of the EMG signal above 160 Hz and the constants are derived from Perry et al. (2001). A muscle was classified as active if the smoothed raw EMG signal was above the threshold level. Subsequently, the active phases were classified as eccentric, concentric or isometric depending on the observed elbow movement and the primary mechanical function of the muscle (i.e. flexion or extension). For example, BBB muscle activity above threshold was classified as concentric when the elbow was being flexed and as eccentric when the elbow was being extended. Above threshold, TBL muscle activity was classified as concentric for elbow extension and as eccentric activity for elbow flexion. If the muscle was active but no change in elbow angle was observed, it was classified as isometric activity. However, this isometric activity was not included in further analysis of this study since the task involved a dynamical movement with accordingly very short relative durations of isometric activity (1.25% of the total muscle activity). The duration of all eccentric and concentric phases was summed and expressed as a percentage of the total movement time (i.e. the movement time of the first two cycles), giving the relative duration of eccentric activity and the relative duration of concentric activity for each muscle.

Statistical analysis

The effect of viewing side and visual feedback condition on the bimanual coupling, EMG intensity and the phases of muscle activity in each arm, was tested using a repeated measurement ANOVA with three within factors: Limb (impaired, less-impaired), Viewing side (view impaired [ViewImp], view less-impaired [ViewLessImp]) and Visual condition (mirror, screen, glass). These analyses were conducted using mean data calculated from the three trials per combination of independent variables. In the event that the sphericity assumption was violated, Greenhouse-Geisser adjustments were applied. Fisher's LSD tests were used for the post hoc analysis, and the level of significance was set at 0.05.

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[2] Only the first two cycles of each trial could be analyzed since some children with SHCP could only fulfil 2 cycles before they adopted a different coordination mode than the one they were instructed to produce. Moreover, for some children the movement time allowed them to complete only 2 cycles within the allocated time of each trial or the hand slipped off the handle at which point the trial had to be terminated.

Results

Bimanual coupling

The CRP did not differ in the three visual conditions (mirror = 6.6° ± 6.3°; screen = 13.2° ± 7.2°; glass = 10.8° ± 7.4°) and the viewing side did not have an effect on the interlimb coupling either (ViewImp = 11.1° ± 6.4° and ViewLessImp = 9.3° ± 7.0°; see Table 2 for values per individual condition). The overall mean was 10.2° ± 6.6°, indicating that the less-impaired arm was the leading limb.

Intensity of the mean neuromuscular activity in BBB and TBL

There were no significant main or interaction effects on the mean neuromuscular activity in BBB and TBL of either Viewing side or Visual condition (see Table 3). This means that the EMG intensity in BBB and TBL did not change as a function of viewing side or the nature of visual feedback. Viewing the impaired arm and its mirror reflection did not result in higher levels of EMG intensity (BBB: 24.1 ± 3.1; TBL: 9.9 ± 1.2) than viewing the less-impaired arm and its mirror reflection (BBB: 21.7 ± 3.6; TBL: 11.2 ± 2.0). Inspection of Table 3 seems to indicate a trend ($F_{2,18} = 2.76$, $P = 0.09$) towards lower intensities of neuromuscular activity in the mirror condition compared with the glass and the screen conditions (especially in the BBB of the less-impaired limb in the ViewLessImp condition). In addition, the mean neuromuscular activity tended to be higher in the impaired than in the less-impaired arm for both the BBB and TBL muscles (BBB: 29.0 ± 4.9 vs. 19.5 ± 3.9; TBL: 14.7 ± 3.3 vs. 8.5 ± 1.1); however, the ANOVA indicated that this effect of Limb was not statistically significant (BBB: $F_{1,9} = 2.29$, $P = 0.17$; TBL: $F_{1,9} = 3.40$, $P = 0.10$).

Relative duration of concentric and eccentric activity in the BBB muscle

No significant main or interaction effects were found for the concentric activity of the BBB muscle (see Table 4). Mirror visual feedback, irrespective of which arm was viewed, did not have an effect on the relative contribution of concentric BBB activity to the execution of the movement in the impaired or less-impaired arm ($F_{2,18} = 0.36$; $P = 0.70$). Additionally, there tended to be more concentric activation in the impaired limb than in the less-impaired limb (25.8 ± 3.9 vs. 17.2 ± 4.4), but this difference was insignificant ($F_{1,9} = 2.74$, $P = 0.13$).

Table 2 Mean and SE values of the continuous relative phase (CRP) in degrees for each visual condition and viewing condition

	ViewImp	ViewLessImp
Mirror	8.1 ± 7.7	5.0 ± 6.6
Screen	17.2 ± 7.1	9.3 ± 8.6
Glass	8.0 ± 6.6	13.6 ± 8.6

Table 3 Mean and SE values of the intensity of mean neuromuscular activity (μ*V*) for the BBB and the TBL muscle of the impaired and the less-impaired limb presented for each viewing condition (ViewImp, ViewLessImp)

	BBB	
	ViewImp	ViewLessImp
Impaired limb		
Mirror	29.9 ± 4.2	27.4 ± 5.7
Screen	27.9 ± 4.2	27.3 ± 5.6
Glass	31.0 ± 6.3	30.6 ± 5.2
Less-impaired limb		
Mirror	18.2 ± 3.8	16.2 ± 3.2
Screen	17.6 ± 3.4	21.3 ± 4.4
Glass	17.5 ± 4.5	26.2 ± 7.2
	TBL	
	ViewImp	ViewLessImp
Impaired limb		
Mirror	12.4 ± 2.2	13.9 ± 3.5
Screen	12.4 ± 2.0	17.3 ± 5.4
Glass	15.4 ± 4.3	16.8 ± 3.9
Less-impaired limb		
Mirror	7.3 ± 1.1	8.4 ± 1.4
Screen	8.8 ± 1.3	8.8 ± 1.4
Glass	6.8 ± 1.1	10.6 ± 1.9

For the eccentric activity of the BBB muscle, a significant main effect of the Limb was found ($F_{1,9} = 7.53$, $P = 0.02$) with the impaired limb having 16.3% more eccentric activity than the less-impaired limb. This effect was accompanied by a three-way interaction between Limb, Viewing side and Visual condition ($F_{2,18} = 4.67$, $P = 0.02$). Figure 2 illustrates this interaction using the difference in eccentric activity between the two viewing sides (i.e. ViewImp and ViewLessImp) for the impaired and less-impaired limb and for each visual condition. This *difference score* was determined by subtracting the eccentric activity in the ViewImp condition from the eccentric activity in the ViewLessImp condition. A negative difference score then indicates lower eccentric activity in the ViewLessImp condition, whereas a positive difference score represents higher eccentric activity in the ViewLessImp condition. Inspection of Fig. 2 and post hoc examination of the three-way interaction indicated that there were no effects of Visual condition or Viewing side

Table 4 Mean and SE values of the eccentric and concentric muscle activity, expressed as a percentage of the total movement, of the Biceps Brachii Brevis (BBB) and the Triceps Brachii Longus (TBL) in the impaired and less-impaired limb for theViewImp (viewing the movement from the impaired side of the body) and ViewLessImp (viewing the movement from the less-impaired side of the body) conditions

	BBB (%muscle activity)			
	Eccentric		Concentric	
	ViewImp	ViewLessImp	ViewImp	ViewLessImp
Impaired limb				
Mirror	34.2 ± 4.9	23.9 ± 6.5	26.6 ± 3.7	26.1 ± 4.2
Screen	30.2 ± 5.5	28.5 ± 7.2	25.7 ± 4.7	22.5 ± 3.6
Glass	28.0 ± 6.1	36.7 ± 6.3	25.1 ± 5.4	28.6 ± 4.1
Less-impaired limb				
Mirror	12.5 ± 4.1	13.2 ± 4.5	16.4 ± 5.1	16.2 ± 4.5
Screen	12.2 ± 4.1	16.3 ± 4.3	17.4 ± 5.0	18.8 ± 4.6
Glass	15.1 ± 5.6	14.5 ± 3.7	16.2 ± 5.3	18.3 ± 5.2

	TBL (%muscle activity)			
	Eccentric		Concentric	
	ViewImp	ViewLessImp	ViewImp	ViewLessImp
Impaired limb				
Mirror	7.3 ± 2.8	11.6 ± 4.2	10.5 ± 3.7	9.9 ± 4.9
Screen	9.1 ± 3.4	11.7 ± 4.0	11.8 ± 3.4	13.5 ± 5.2
Glass	10.8 ± 4.6	13.0 ± 4.8	12.7 ± 4.5	13.0 ± 4.7
Less-impaired limb				
Mirror	3.4 ± 1.6	4.9 ± 2.3	1.7 ± 0.7	3.8 ± 1.4
Screen	5.2 ± 1.8	3.2 ± 1.2	4.3 ± 1.5	5.7 ± 2.0
Glass	2.2 ± 1.5	8.3 ± 2.6	1.8 ± 1.2	8.8 ± 3.0

on the eccentric activity of the less-impaired arm. For the impaired arm, however, mirror visual feedback from the impaired arm resulted in 10.3% more eccentric activity than mirror visual feedback from the less-impaired arm ($P = 0.007$). Furthermore, a significant effect of Viewing side was also present in the glass condition, where looking from the less-impaired side resulted in more eccentric activity than looking from the impaired side (mean difference score = 8.7%, $P = 0.02$). Viewing side did not have an effect on the eccentric activity of the BBB in the screen condition. Finally, focusing on the differences in eccentric activity between the visual conditions (see Table 4), it was found that mirror visual feedback of the less-impaired arm resulted in less eccentric activity in the impaired arm than the glass condition when viewing from the same side (mean difference = 12.8%, $P = 0.001$). In addition, the glass condition was performed with more eccentric activity in the impaired arm than the screen condition (mean difference = 8.2%, $P = 0.02$).

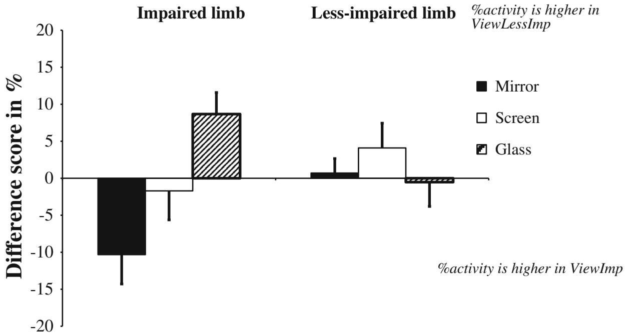

Fig. 2 Difference scores of the relative duration of eccentric activity (in *percentage*) in the BBB muscle of the impaired (*left side* of the figure) and the less-impaired limb (*right side* of the figure) for the mirror (*black bars*), screen (*white bars*) and glass (*dashed bars*) condition. A positive difference score means that the eccentric activity is higher in the ViewLessImp compared with the ViewImp condition, and a negative difference score means that the eccentric activity is lower in the ViewLessImp condition compared with the ViewImp condition

Relative duration of concentric and eccentric activity in the TBL muscle

For the concentric activity of the TBL muscle, a significant interaction effect between Limb and Viewing side was found ($F_{1,9} = 10.47$, $P = 0.01$; see Table 4). The concentric activity in the impaired limb was larger than in the less-impaired limb for both the ViewImp and the ViewLessImp condition (mean difference = 8.56 and 4.56%, respectively). Furthermore, viewing from the less-impaired side resulted in longer durations of concentric activity in the less-impaired limb than viewing from the impaired side, irrespective of the visual condition (mean difference = 3.49%).

For the eccentric activity of the TBL, no effect of Limb, Visual condition or Viewing side was found.

Discussion

This study investigated the effect of mirror visual feedback from the impaired arm (‘compromised’) compared with the mirror visual feedback from the less-impaired arm (‘uncompromised’) on the interlimb coupling and the neuromuscular control during a bimanual coordination task in children with SHCP. In doing so, we wanted to determine whether previously found effects of the mirror box illusion in these children (Feltham et al. 2010a, c) were the result of the mirror and the related perception of visual symmetry per se or of the illusion that the impaired arm appears to move with less jerk and in synchrony with the less-impaired arm. While the former would mean that ‘compromised’ as well as ‘uncompromised’ mirror visual feedback can trigger an improvement of the bimanual coupling and/or the neuromuscular activation, the latter can only be elicited by ‘uncompromised’ mirror visual feedback.

The CRP, which gives an indication of the nature of the bimanual coupling during this task, i.e., the synchronicity of the two limbs, indicates that the less-impaired arm was ‘leading’ the impaired arm across all conditions. This is in congruence with earlier studies on bimanual coordination in typically developing children (Pellegrini et al. 2004) and adults (e.g. Amazeen et al. 1997; Stucchi and Viviani 1993; Treffner and Turvey 1995). The asynchrony of approximately 10° falls within the higher range of previously reported values in children with SHCP (Feltham et al. 2010a: −0.3°; Volman et al. 2002: −5° to 9°), but is still acceptable given the unilateral impairment of the children. Note that the phase lag between the two hands may indicate that the movement of the lagging impaired hand may be guided by visual feedback from the less-impaired hand. However, the CRP did not change as a function of visual condition or viewing side, which suggests that the bimanual coupling is clearly not solely governed by a visual feedback mechanism and that processes relying on central representations of action do contribute to the coupling as well (addressed below).

It thus seems that mirror visual feedback did not influence the interlimb coupling, and there was no difference between ‘compromised’ and ‘uncompromised’ mirror visual feedback. Interestingly, however, the mirror did have an effect on the neuromuscular activity required to perform the task. This suggests that, although the movement performance itself remained the same, the muscular effort responsible for this movement did change in response to the available visual information. Our results demonstrate that mirror visual feedback led to a reduction in eccentric BBB activity in the impaired arm compared with the glass condition, and importantly, this effect was exclusive to ‘uncompromised’ mirror visual feedback, i.e., viewing the less-impaired arm and its mirror reflection (ViewLessImp). In the impaired arm, mirror visual feedback of the less-impaired arm appears to have the capacity to improve the neuromuscular efficiency by reducing the disproportionally high eccentric activity. The finding that ‘compromised’ mirror visual feedback did not elicit a similar effect shows that the mirror effect in children with SHCP is not just a response to the visual symmetry, but is also dependent on the type of visual information generated by the mirror. The latter nuances the findings of Franz and Packman (2004) who found that mirror visual feedback enhanced the bimanual coupling (i.e. similarity in range of motion of the two hands) in typical adults, irrespective of viewing mirror feedback from the left or the right hand. However, unlike in typical adults, in children with SHCP, the nature of mirror visual feedback from the left and right hand is qualitatively different, which might explain the apparent discrepancy between the two studies.

The finding from the present study that mirror visual feedback of the impaired arm has the opposite effect of ‘uncompromised’ apparent symmetrical motion in children with SCHP qualifies the findings of Feltham et al. (2010c) who only looked at the effect of mirror feedback from the less-impaired arm. We demonstrated that the favourable results (i.e. the reduction in eccentric BBB activity in the impaired arm) are not just due to the visual perception of apparent bimanual symmetry per se. Instead, children with SHCP appear to benefit specifically of mirror visual feedback from the less-impaired arm, which seems to be in line with the notion of Ramachandran (2005). Ramachandran hypothesised that mirror visual feedback may assist the central control of movement in people with unilateral motor problems by restoring the congruence between disrupted sensory information and the central motor command signals. According to this view, the information provided by the mirror could assist in the neuromuscular control of the movement by replacing conflicting visual feedback of the impaired limb with feedback that is in accordance with the intended movement (i.e. ‘uncompromised’ visual feedback of the less-impaired limb). By showing that the mirror effect on motor performance in children with SHCP is specifically related to mirror visual feedback of the less-impaired arm, the current study provides a valuable contribution to the discussion about the underlying mechanisms of this effect. Nevertheless, the actual neural underpinnings will only be revealed using advanced neuroimaging techniques. In addition, it may be surprising that a short exposure to the mirror already induces these effects on the neuromuscular activity and future studies should examine the impact of longer exercise or interventions with mirror feedback. Related to this issue is the fact that no (major) effect of the mirror was observed on the bimanual coupling or neuromuscular measures such as the intensity of mean neuromuscular activity, the eccentric activity in the TBL muscle and concentric activity in the BBB muscle. Furthermore, we cannot exclude the limited number of trials (three per condition) and the large age range of the participants to affect the precision and generalisation of the results. The precision of the measurement might be enhanced with larger number of trials, but in the current study, it was high enough to reveal significant differences between the conditions. One can expect that a larger number of trials will enhance the actual results but one must also consider that the limited attention span and fatigability of the participants with cerebral palsy might interfere. Considering that the present study used a repeated measures design each participant was his own control and the variability that the large age range may have introduced was nevertheless small enough to show a significant effect of the experimental conditions. While we did not anticipate an age effect, we cannot exclude it and suggest that this should be further investigated.

In conclusion, this study provided more insight into the effects of mirror visual feedback in children with SHCP. We showed that the effects found by Feltham et al. (2010a, c) on neuromuscular activity and bimanual coordination are likely not caused by the perception of two symmetrically moving limbs per se. Instead, for an increase in neuromuscular efficiency of bimanual movement (i.e. a decrease in excessive eccentric activity in the arm flexors), children with SHCP require mirror visual feedback of the ('unaffected') less-impaired limb.

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Visual feedback of the non-moving limb improves active joint-position sense of the impaired limb in Spastic Hemiparetic Cerebral Palsy

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Mirror visual feedback

A B S T R A C T

This study examined the active joint-position sense in children with Spastic Hemiparetic Cerebral Palsy (SHCP) and the effect of static visual feedback and static mirror visual feedback, of the non-moving limb, on the joint-position sense. Participants were asked to match the position of one upper limb with that of the contralateral limb. The task was performed in three visual conditions: without visual feedback (no vision); with visual feedback of the non-moving limb (screen); and with visual feedback of the non-moving limb and its mirror reflection (mirror). In addition to the proprioceptive measure, a functional test [Quality of Upper Extremity Skills Test (QUEST)] was performed and the amount of spasticity was determined in order to examine their relation with proprioceptive ability. The accuracy of matching was significantly influenced by the distance that had to be covered by the matching limb; a larger distance resulted in a lower matching accuracy. Moreover it was demonstrated that static (mirror) visual feedback improved the matching accuracy. A clear relation between functionality, as measured by the QUEST, and active joint-position sense was not found. This might be explained by the availability of visual information during the performance of the QUEST. It is concluded that static visual feedback improves matching accuracy in children with SHCP and that the initial distance between the limbs is an influential factor which has to be taken into account when measuring joint-position sense.

1. Introduction

Cerebral palsy (CP) is, with an incidence of 2–2.5 per 1000 living births, one of the most common childhood disorders (Lin, 2003). The condition is caused by damage to the brain and/or pyramidal tract and depending on the location of the lesion and the clinical outcome of the damage, different forms of CP are distinguished. In Spastic Hemiparetic Cerebral Palsy (SHCP) the damage is limited to one side of the brain leading to impaired control of muscle tone and spasticity in the lower and upper limbs on the contra-lesional side of the body (Albright, 1996). Although SHCP is classed as a unilateral condition, recent studies have highlighted that children with SHCP have motor difficulties beyond their unilateral deficits. The spasticity of the impaired limb limits the performance of bimanual tasks and evidence suggests mild motor impairments in the unaffected limb as well (Steenbergen & Meulenbroek, 2006).

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Impairments as spasticity are often accompanied by disturbances in proprioception (Cooper, Majnemer, Rosenblatt, & Birnbaum, 1995; Odding, Roebroeck, & Stam, 2006). Proprioception is a complex somatosensory modality that consists of two components: kinaesthesia and joint-position sense. Kinaesthesia is defined as the sense of limb movement whereas joint-position sense is referred to as static limb position (Goble, Lewis, Hurvitz, & Brown, 2005). Proprioception plays a major role in performing and controlling movements including updating motor plans based on e.g. monitoring movement execution through comparison of predicted and actual movement outcomes (Goble, 2006). A number of studies have demonstrated that the proprioceptive ability of children with SHCP is impaired (Goble, Hurvitz, & Brown, 2009; Wann, 1991; Wingert, Burton, Sinclair, Brunstrom, & Damiano, 2009), and there are indications that the impaired limb has a poorer proprioception than the less-impaired limb (Goble, Hurvitz, et al., 2009; Wingert et al., 2009). Furthermore, in addition to the differences in proprioception between the limbs, Goble, Hurvitz, et al. (2009) also found a difference in proprioceptively guided matching tasks between individuals with a left brain lesion and individuals with a right brain lesion. In individuals with a right hemispheric lesion (RHL) the proprioceptive ability was more impaired than in individuals with a left hemispheric lesion (LHL). Goble's findings can be supported by neuroimaging studies which showed that the right hemisphere is more activated during the performance of a proprioceptive task (Naito et al., 2005).

Although proprioception is impaired in individuals with SHCP, they are still able to sustain a certain level of movement accuracy, implying that visual information is used to attain this movement accuracy (van Roon, Steenbergen, & Meulenbroek, 2005). Indeed, studies by Wingert et al. (2009) and Wann (1991) on individuals with CP demonstrated that vision of the moving upper limb improved the performance on the joint-position task compared to a situation in which no visual feedback of the moving upper limb was available. However, Wann (1991) also showed that visual information of the non-moving hand did not improve movement accuracy in a joint-position sense task for individuals with bilateral CP. According to Wann (1991) this suggests that individuals with bilateral CP have difficulties encoding the visual and proprioceptive information into a common reference frame. However, the possibility that visual feedback of the non-moving limb might afford a reference frame for the proprioceptive information of the moving limb has not been investigated in individuals with hemiplegia. One of the explanations for the problems in encoding proprioceptive and visual information that Wann (1991) presents is that the cortical damage may have destroyed the neural structures that are necessary for egocentric mapping. This might indeed be the case for diplegic patients, but children with hemiplegia have a lesion in one hemisphere. It therefore might be possible that patients with hemiplegia are able to encode proprioceptive and visual information into a common reference frame. Therefore, the present study will examine the effect of visual feedback of the non-moving limb on the contralateral matching performance of the moving limb in this population. Given the asymmetry in proprioception in hemiplegia but also given the fact that only one hemisphere is damaged, it can be expected that the visual and proprioceptive information of the non-moving (less-impaired) upper limb might be integrated into one egocentric reference frame for the moving (impaired) upper limb (Jeannerod, 1986; von Hofsten & Rosblad, 1988; Wann, 1991), facilitating the contralateral matching in comparison to a situation in which no visual feedback is available.

In addition to the effect of visual information of the non-moving limb, the current study investigates the effect of mirror visual feedback of the non-moving limb on the movement accuracy during a contralateral matching task in children with SHCP. Mirror visual feedback has been demonstrated to have a positive effect on the bimanual coordination and neuromuscular activity in children with SHCP (Feltham, Ledebt, Bennett, et al., 2010; Feltham, Ledebt, Deconinck, & Savelsbergh, 2010). However, Holmes and Spence (2005) showed that manipulating the position of the moving hand (behind the mirror) influenced unimanual reaching movements in TD adults negatively. They suggested that this was the result of an integration of visual and proprioceptive information of the non-moving limb which caused a bias in the felt initial position of the moving hand. It can thus be hypothesized that providing mirror visual of the non-moving (less-impaired) upper limb (thus seeing two non-moving upper limbs), would deteriorate the contralateral matching performance of the impaired upper limb in children with SHCP. In the forthcoming, visual feedback of the non-moving limb will be referred to as static visual feedback and visual feedback of the moving limb will be referred to as dynamic visual feedback. Mirror visual feedback of the non-moving limb will be referred to as static mirror visual feedback.

Literature on the relationship between impaired proprioception and other impairments in CP as well as the relationship with the activity level is scarce. The relationship with spasticity was assessed in the study of Chrysagis, Skordilis, Koutsouki, and Evans (2007) who showed that an increase in spasticity was related to a decreased performance on an active joint-position sense task. Accordingly, Tardieu, Tardieu, Lespargot, Roby, and Bret (1984) stated that spasticity causes disturbances in the muscle spindle functioning leading to inappropriate kinaesthetic feedback (Chrysagis et al., 2007). However, the relationship between arm/hand functionality and joint-position sense has, to the best of our knowledge, not been examined yet. In order to get more insight into the influence of spasticity on joint-position sense and to clarify the impact of an impaired joint-position sense on daily functioning, the current study will investigate these two relationships.

In general, the present study aimed to get more insight into the proprioceptive impairments of the impaired and the less-impaired upper limb in children with SHCP. We assessed the role of static visual feedback and static mirror visual feedback on joint-position sense of the upper limbs using three different visual conditions: a no vision condition without any visual feedback of both limbs, a screen condition in which only the non-moving reference limb was visible (static visual feedback) and a mirror condition in which the non-moving reference limb was visible and its reflection in the mirror (static mirror visual feedback). It was hypothesized that static visual feedback of the less-impaired limb would improve the movement accuracy of the impaired limb compared to the situation without visual feedback. In addition, it was expected that static mirror visual feedback would create a conflict situation between the visual and proprioceptive feedback which would result in a deteriorated performance.

Furthermore, the current study aimed to examine the relationship between one of the main impairments in CP, spasticity, and the impaired proprioception in CP, and between the impaired proprioception and the arm/hand functionality. It was hypothesized that a higher degree of spasticity would be related to an impaired joint-position sense which would in turn be linked to a deteriorated arm/hand functionality. Finally, differences in joint-position sense impairment between left and right hemispheric brain lesions were examined. Following the findings of Goble, Hurvitz, et al. (2009) it was hypothesized that individuals with a right hemispheric lesion would have a more deteriorated joint-position sense than individuals with a left hemispheric lesion.

2. Methods

2.1. Participants

14 children with SHCP participated in the study (age 12.6 ± 1.95). 6 children had a right hemispheric lesion and 8 children had a left hemispheric lesion. Individual participant characteristics can be found in Table 1. None of the participants had any neuromuscular disorder other than SHCP, pain in either of the upper limbs, visual neglect, visual impairments not corrected to normal, mental retardation, or received a treatment with Botulinum toxin in either of the arms in the past six months preceding the measurement. The children with SHCP were recruited through the Dutch society for children with a physical handicap and their parents (BOSK). Participants' parents provided written informed consent prior to testing. All procedures were approved by the institutional research ethics committee and in accordance with the Declaration of Helsinki.

2.2. Measures of functionality

Before the actual start of the experiment different measures were performed to examine the participants' body functions. Additional information about the child's disorder was obtained from a general questionnaire, filled in by the parents, with questions about e.g. the cause and severity of the disorder and limitations the child faces in daily life. In addition, the parents were asked to fill in The Functional Independence Measure (WeeFIM). The WeeFIM measures the functional abilities in activities of daily life like the ability to feed, dress and bathe (Ottenbacher, Hsu, Granger, & Fiedler, 1996). For the current study only the WeeFIM motor items were used.

Grip strength was determined for each upper limb, using a hand-held dynamometer measuring the average of three maximum voluntary contractions in kilograms (JAMAR, digital hand dynamometer, Clifton, USA).

The Quality of Upper Extremity Skills Test (QUEST) (DeMatteo et al., 1992) was performed to qualify the functional ability of the arms and hands of each participant. This test consists of 7 domains, however for this study only the parts about "Dissociated movements" (part A) and "Grasps" (part B) were conducted since these two domains were specifically related to the task the children had to perform during the measurement. The QUEST is validated for children between 18 months and 8 years of age (DeMatteo et al., 1992). However, although the mean age of our population is 12.6 years it was still chosen to use the QUEST since this test is more extensive than other tests that measure the functioning of the upper limbs. Based on the items of the two included parts of the QUEST and the related scoring criteria we calculated separate scores for the impaired and the less-impaired limb. A higher score on this selection of QUEST items represents a better functionality. Table 2 presents the individual QUEST scores.

In addition to the QUEST, the Manual Ability Classification System (MACS) level was determined. The MACS describes how children use their hands during object handling and their need for assistance to perform manual skills in everyday life

Table 1
Subject characteristics.

Participant	Age (years)	Sex	Side brain lesion	Grip strength impaired/less-impaired limb (kg)	TSelbow$_{\text{(flex-ext)}}$/ TSwrist$_{\text{(flex-ext)}}$[a]	WeeFIM/MACS	Aetiology
1	13.4	M	Right	11.7/52.3	3-1/2-2	78/3	O_2 shortage during birth
2	10.5	M	Right	4.0/44.0	3-1/3-0	88/3	Thrombosis
3	10.8	M	Right	12.3/30.0	2-1/1-0	91/2	Unknown
4	14.5	M	Right	7.3/52.3	2-2/2-0	62/3	Schizen cephaly
5	13.6	M	Right	14.7/52.0	2-2/0-0	91/2	Cerebral infraction
6	10.8	F	Right	4.7/22.0	2-1/0-0	52/3	Cerebral haemorrhage
7	12.1	F	Left	2.0/63.7	2-0/2-1	91/3	Thalamus infarction at birth
8	15.5	M	Left	60.3/105.7	2-0/0-0	76/1	Unknown
9	9.3	M	Left	23.3/49.7	2-0/0-0	91/1	Cerebral infarction
10	13.1	F	Left	25.0/69.7	2-2/0-0	91/2	Cerebral infarction
11	14.4	M	Left	0.0/104.0	2-0/0-0	81/2	Cerebral haemorrhage
12	12.5	M	Left	0.0/62.0	2-2/2-0	59/3	Cerebral infarction
13	14.3	M	Left	13.6/101.3	2-2/1-0	71/3	Unknown
14	10.6	M	Left	24.7/69.0	0-1/0-0	87/2	O_2 shortage during birth

[a] Tardieu score (TS) is only of the impaired limb. (flex/ext) are separate scores for flexion and extension.

Table 2
QUEST scores; total score and scores of part A (dissociated movements) and part B (grasps) for each limb.

Participant	Total score	Part A impaired limb	Part A less-impaired limb	Part B impaired limb	Part B less-impaired limb
1	72.2	60.0	99.2	86.7	100
2	51.1	57.0	100	50.0	80.0
3	82.5	86.6	99.1	81.7	88.3
4	65.3	72.5	100	60.0	80.0
5	68.5	66.5	100	73.3	90.0
6	52.6	64.8	99.2	48.3	85.0
7	77.4	71.5	100	85.0	100
8	96.4	98.4	100	96.7	98.3
9	95.9	99.2	100	93.3	93.3
10	81.7	78.1	100	86.7	100
11	55.2	54.8	100	60.0	100
12	51.4	54.7	100	55.0	93.3
13	63.0	70.7	98.4	65.0	95.0
14	85.1	77.3	98.4	95.0	95.0

(Carnahan, Arner, & Hagglund, 2007). The severity of performance limitation and the degree of required assistance increases for each MACS level from I to V. The MACS levels and their specifications are depicted in Table 3. The performances of the QUEST were recorded with a digital video camera (JVC Hard disk Camcorder, HDD F1.2, GZMG40E) in order to score the performances afterwards. Two experimenters analyzed the video tapes independently. The inter-rater reliability was high ($r = 0.92$, $p < 0.001$).

The degree of spasticity was determined by a qualified physiotherapist using the Tardieu scale. The assessment involved passive movement of the arm in the sagittal plane, first as slow as possible and second as fast as possible, while the child was seated on a chair with the knees bend in 90°. The physiotherapist quantified the spasticity of the arm muscles (biceps brachii brevis, triceps brachii longus, flexors and extensors of the wrist) during the fast velocity stretch according to the criteria of muscle reaction for grades 0–3. The definition of each grade is depicted in Table 4. The Tardieu score averaged for the biceps and the triceps was further used for analysis.

2.3. Procedures

The child was seated on a height adjustable chair at a height adjustable table with the knees 90° flexed. Joint-position sense was assessed using a custom made device consisting of two handles, each on a separate track fixed to a horizontal panel. The tracks were 20 cm apart, parallel to each other, and perpendicular to the medio-lateral axis of the trunk. The handles could be moved within a range of 56 cm. The children were positioned such that the centre of the body was located in between the two tracks, and with the beginning of the track at 15 cm from the upper body. The position of the handles was recorded outside the wooden device using one Optotrak unit with three infrared cameras (3020 Optotrak, Northern Digital Inc., Waterloo, Canada). The experimental setup is depicted in Fig. 1.

Before the start of the measurement, the maximum reaching distance of the impaired arm was determined (MRD) in order to scale the different matching positions across subjects. MRD was the distance from the start of the track to the position of the handles when the elbows were extended as far as possible without bending the trunk forward. If a participant was unable to grip the handle due to physical impairment, the experimenter placed the hand on top of the handle. All participants were able to hold the handles during the whole experiment.

Table 3
Description for each MACS level.

MACS level	Description
I	Handles objects easily and successfully.
II	Handles most objects but with somewhat reduced quality or speed of achievement.
III	Handles objects with difficulty; needs help to prepare or modify activities.
IV	Handles a limited selection of easily managed objects in adapted situations.
V	Does not handle objects and has severely limited ability to perform even simple actions.

Table 4
Tardieu scale scoring the quality of muscle reaction to stretch.

0	No catch, no resistance.
1	Light resistance without clear catch.
2	Clear catch followed by a release.
3	Clear catch, no release.

Fig. 1. Experimental setup during the no vision (left panel), screen (middle panel), and mirror (right panel) condition.

The active joint-position sense task required participants to match the position of one limb (reference limb), fixed at 25%, 50%, or 75% of the MRD, by actively moving the other limb (matching limb). The task was performed with either the less-impaired limb or the impaired limb and the matching started at the MRD (distal) or at the beginning of the track (proximal). The matching task was performed in three different visual conditions: a no vision condition (both hands were not visible), a screen condition (only the reference hand was visible), and a mirror condition (only the reference hand was visible and its reflection in the mirror). The position of the reference limb (3), the matching limb (2), the start position of the matching limb (2), and the visual conditions (3) resulted in 36 trials. The conditions were randomly presented to the participant but all trials with the same matching limb were kept together even as the trials within one visual condition. Prior to data collection 3 practice trials were conducted to familiarize the participant with the test setup. In order to keep the children motivated they were told that the better their performance the more points they could get. At the end of the experiment they could trade their points for a small gift.

2.4. Data analysis

A custom made Matlab program (The Mathworks, Inc.) was used to determine the absolute difference (error) between the position of the reference limb and the position of the matching limb at the end of the movement. The end of the movement was indicated by visual inspection (see Fig. 2).

Goble, Coxon, Wenderoth, Van Impe, and Swinnen (2009) stated that several studies that measured proprioceptive acuity found larger errors for the matching of targets farther from the body in contrast to targets closer to the body. However, in these studies the starting position was the same for all trials and hence it can be argued that the distance that has to be covered by the matching limb is the influencing factor instead of the position relative to the body. This idea is supported by Smorenburg, Ledebt, Deconinck, and Savelsbergh (submitted for publication) who found larger errors when the distance covered by the matching limb was larger. Therefore the current study combined the two starting positions (distal, proximal)

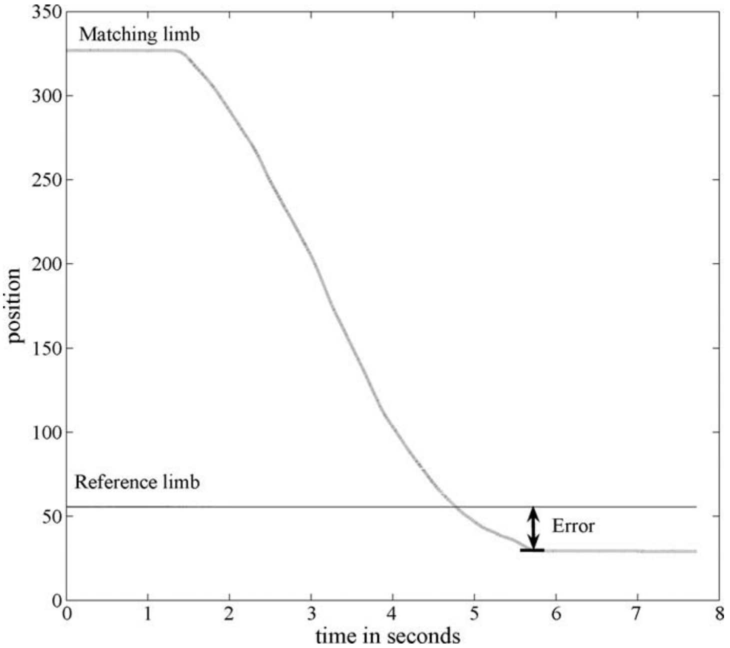

Fig. 2. Example of a movement pattern. The arrow indicates the distance between the limbs at the end of the movement.

of the matching limb and the three positions of the reference limb (25%, 50%, 75% of the MRD) into three distances that had to be covered by the matching limb (small, medium, large).

2.5. Statistical analysis

A repeated measurement ANOVA was performed with Distance (small, medium, large), Matching limb (impaired, less-impaired) and Visual condition (mirror, screen, no vision) as within factors. Lesion side [left hemispheric lesion (LHL), right hemispheric lesion (RHL)] was taken as between factor. If the sphericity assumption was violated, Greenhouse Geisser adjustments were made. Post hoc comparisons for the interaction effects were performed with the Fishers' LSD test.

2.6. Correlations

Correlations were calculated using the Pearson's correlation coefficient (r). For the correlations with the Tardieu scale, Spearman's correlation coefficient was used (r_s).

3. Results

3.1. Matching accuracy

The accuracy of active matching was significantly influenced by Distance ($F_{(1.2,14.1)} = 8.71$, $p = 0.008$), showing a general trend that the absolute error became gradually larger with larger matching distances. Other main effects were absent, but all factors were involved in second order interactions (Hand × Distance: $F_{(2,24)} = 3.99$, $p = 0.032$; Visual condition × Distance:

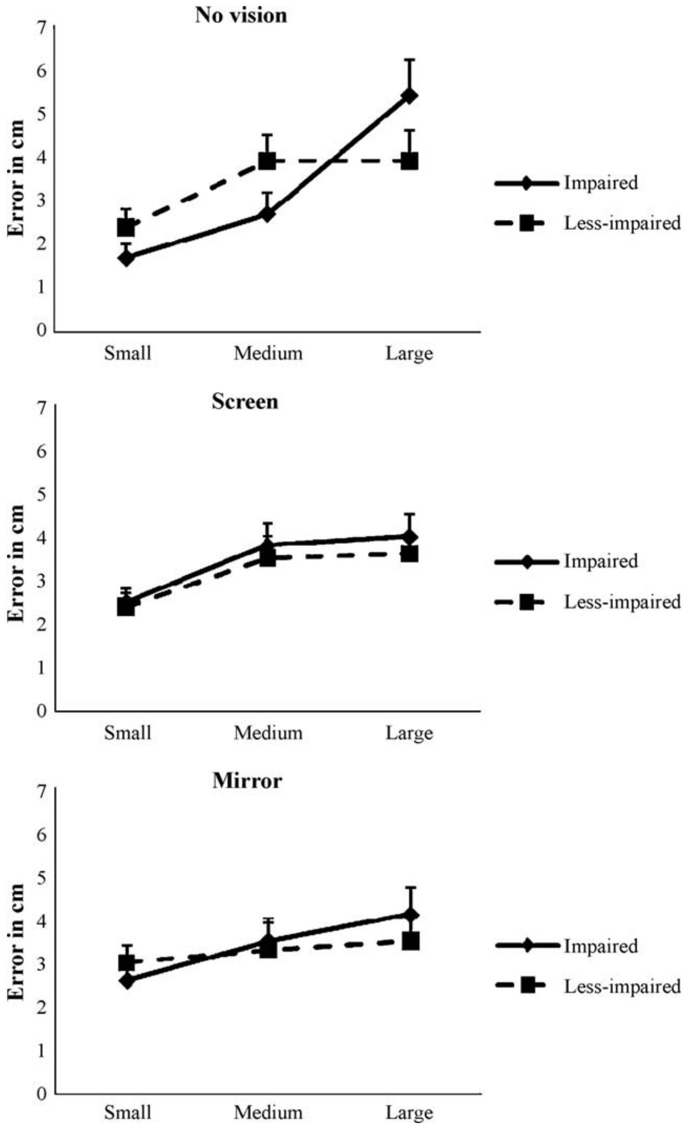

Fig. 3. Hand by Distance by Visual condition (no vision, screen, mirror) interaction.

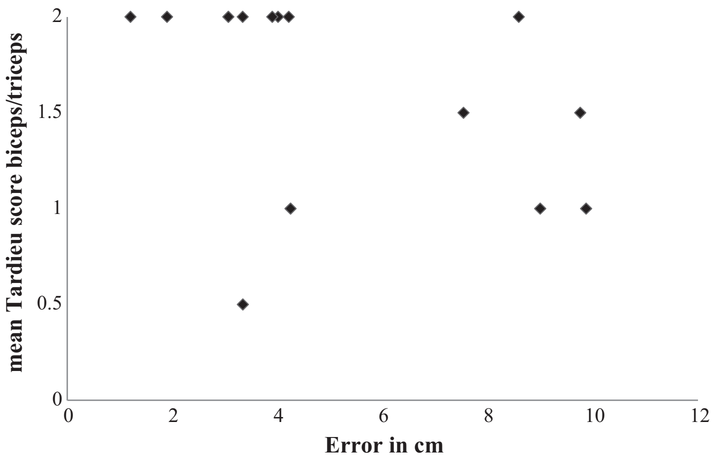

Fig. 4. Correlation between the Tardieu score averaged for the biceps and triceps and the error on the active joint-position sense task of the impaired limb in the no vision condition for the large distance.

$F_{(4,48)} = 3.81$, $p = 0.009$) and a third order interaction (Hand × Distance × Visual condition: $F_{(4,48)} = 3.26$, $p = 0.019$) (see Fig. 3). Fig. 3 reveals similar trends for all visual conditions in the less-impaired limb and the screen and mirror conditions in the impaired limb. In accordance with the main Distance-effect smaller errors were made in the small distance condition, except for matching with the less-impaired limb in the mirror condition where no significant differences between distances were found. The differences between the two limbs and between the visual conditions were related to the deviant profile of the no vision condition for the impaired hand. Matching large distances with the impaired limb without visual information resulted in significantly larger errors than in the mirror or screen condition. In addition, the impaired limb showed a similar or larger error as the less-impaired limb with exception of the medium matching distance in the no vision condition. Matching with the impaired limb in this condition (medium, no vision) yielded larger errors than for the less-impaired limb, whereas the latter was less accurate than the impaired limb in the large distance, no vision condition. Finally, no differences in accuracy of active matching were found between LHL and RHL.

3.2. Functionality (QUEST) and spasticity

3.2.1. QUEST vs. active joint-position sense

A significant correlation was revealed between the QUEST part A (dissociated movements) of the impaired limb and the error on the active joint-position sense task of the impaired limb in the screen condition for the large distance ($r = -0.70$, $p = 0.006$).

3.2.2. QUEST for left- and right hemispheric lesions

The QUEST score part A (dissociated movements) and the QUEST score part B (grasps) of the impaired upper limb were not significantly different between the LHL and the RHL group. Moreover, for the less-impaired limb no difference between the two groups was revealed for the QUEST score part A, but for the QUEST score part B the RHL group had a higher score than the LHL group (mean difference = 9.65, $p = 0.006$).

3.2.3. Spasticity vs. active joint-position sense

A significant correlation between the mean Tardieu score of the biceps and the triceps and the absolute error on the active task was found. A higher Tardieu score was related to a smaller error of the impaired limb in the no vision condition for the large distance ($r_s = -0.54$, $p = 0.047$). This relation is depicted in Fig. 4.

4. Discussion

The current study aimed to get more insight into the integrity of proprioception in the impaired and less-impaired limb in children with SHCP. In an active joint-position sense task, different visual conditions were used in order to investigate the effect of static visual feedback and static mirror visual feedback on joint-position sense. In addition, the relation between joint-position sense and spasticity and joint-position sense and arm/hand functionality was investigated. Finally, following the findings of Goble, Hurvitz, et al. (2009) we examined differences in joint-position sense between individuals with a right hemispheric lesion and individuals with a left hemispheric lesion.

A general finding in this study was that the position of the reference limb could be matched with greater accuracy when the distance to be covered was smaller, irrespective of which limb was used to match and irrespective of the initial position of the reference limb (in the proximity of the body or further away). This finding is in agreement with previous results in typically developing children (Goble & Brown, 2008; Goble, Lewis, & Brown, 2006) and children with SHCP (Smorenburg et al., submitted for publication).

A physiological phenomenon that may explain the larger absolute errors for longer reaching or matching distances is the signal-dependent noise on a motor command. According to this principle the variance of the noise on neural control signal increases with the size of the signal (Harris & Wolpert, 1998). This would suggest that for larger distances, requiring the generation of a larger command signal, the variance of noise becomes larger, which will hamper the accurate matching of the upper limbs. In addition to this physiological explanation, it is assumed that factors associated with daily functioning may play a role in the distance-effect, especially when considering the matching task used in the current study. Goble et al. (2005) suggested that the improvements in the acuity of joint-position sense when comparing children and adolescents are partly the result of experience-driven processes. Our daily movement repertoire is diverse, but with respect to grasping and reaching movements the range of motion is typically kept relatively small, which may lead to a distance-specific specialization of proprioception. In this respect it is interesting to note that in the current experiment the error score was highest when matching large distances with the impaired arm. Due to the spasticity, which tends to shorten the muscles leading to partial immobility of this arm (Love et al., 2001), children with SHCP might avoid using the arm for tasks involving larger ranges of motion. This substantial in absolute error increase for the large distance condition was absent when matching with the less-impaired arm. Although a better acuity of this less-impaired arm can be expected, this finding is still remarkable because the contralateral matching task involves the utilization of afferent proprioceptive information from both the reference (impaired) and the matching (less-impaired) arm.

Comparison of the error score across visual conditions indicates that static visual feedback of the reference limb has the capacity to improve joint-position sense, in particular when matching large distances with the impaired arm. This finding is in contrast to those of Wann (1991) who found that a group of children with mixed diagnoses of CP did not benefit from visual information of the reference limb and target in a similar matching task. Wann (1991) showed that the performance of the children with CP for tasks requiring crossmodal matching (between sensory modalities, i.e. vision and proprioception), was lower than in all other conditions where intramodal matching was possible (within one sensory modality). It was concluded that CP was associated with a reduced ability to generate an egocentric frame of reference needed for accurate mapping between sensory modalities. It is important to note that the children participating in Wann's study all suffered bilateral damage to the brain (diplegia and quadriplegia). Our results then imply that in children with unilateral damage to the brain, crossmodal mapping is not disturbed to a similar extent as in diplegic and quadriplegic patients, and still allows the encoding of sensory signals into a common egocentric frame of reference. The beneficial effect of vision in a situation where spasticity compromises matching acuity most (large distance matching with impaired hand), suggests that joint-position sense in children with SHCP seems to be affected by a distortion of the physiological function of the somatosensory organs, rather than by a deficit in higher sensory motor function. Our finding that static visual feedback of the less-impaired limb improves the matching accuracy might potentially be interesting for therapeutic interventions in order to improve the joint-position sense of the impaired limb. If training with static visual feedback of the less-impaired limb can improve the joint-position sense of the impaired limb, this might have implications for the daily functioning of the children. The focus nowadays is primarily on improving motor behaviour by practicing, but since proprioception is an important factor in movement control, this might be another angle of approach in order to improve daily functioning in children with SHCP.

Despite the beneficial effects of static visual feedback, no detrimental effects of static mirror visual feedback were found. Based on the findings of Holmes and Spence (2005) it was expected that static mirror visual feedback would deteriorate the matching accuracy, especially of the impaired limb. However, Holmes and Spence (2005) showed also that a longer exposure time to the mirror resulted in larger errors. The short exposure time in the current study might explain why we did not find an effect of the mirror in the active joint-position sense task. Moreover, in general, proprioceptive information is more reliable under active than under passive conditions. It can be expected that perceived hand position will be less affected by (discrepant) mirror visual feedback in an active compared to a passive condition (Chokron, Colliot, Atzeni, Bartolomeo, & Ohlmann, 2004; Holmes & Spence, 2005; Van Beers, Wolpert, & Haggard, 2002). It is therefore suggested to examine the differences in mirror effect between an active and a passive joint-position sense task.

Based on the study of Goble, Hurvitz, et al. (2009) we expected that differences in joint-position sense between the upper limbs and the effects of visual information would be different for individuals with a left hemispheric lesion and individuals with a right hemispheric lesion, but in the present study no effect of lesion side was found. Differences in task (ipsilateral remembered vs. contralateral matching) between our study and the study of Goble, Hurvitz, et al. (2009) might have caused these discrepant findings. Moreover, in both studies no specific information about the location of the brain lesion is present which makes it difficult to draw clear conclusions. However, the current study examined the functional level of the participants by means of the QUEST, which might shed a light on the severity of the condition. It was shown that participants with LHL and participants with RHL had the same mean QUEST scores for the impaired side of the body. Although both groups in the study of Goble, Hurvitz, et al. (2009) had similar spasticity scores, no information about the functional level was available. Without this information it is impossible to determine whether differences in joint-position sense between individuals with LHL and RHL are actually caused by the side of the lesion or by other factors related to the severity of the condition.

Finally, we looked at the relation between spasticity and joint-position sense and between arm/hand functionality and joint-position sense. One significant correlation between spasticity and joint-position sense was found. However, a close look at the significant correlation shows that seven individuals with a mean Tardieu score of 2 had a relative small error. The other seven participants showed a more scattered distribution. Hence it can be argued that this is not a clear-cut relationship. It is possible that the participants adapted their movement velocity in order to minimize the effect of their spasticity. Since the Tardieu scale is determined at a (fast) speed by the physiotherapist, it is plausible that this speed does not match with the movement speed during the active task. The current findings are in contrast with the findings of Chrysagis et al. (2007) who found that a higher degree of spasticity was related to a more deteriorated joint-position sense. However, Chrysagis et al. (2007) used the Modified Ashworth Scale (MAS) to determine the degree of spasticity whereas we used the Tardieu scale. Although both scales are frequently used as clinical measure, the inter-rater reliability and test–retest reliability are better for the Tardieu than for the MAS (Fosang, Galea, McCoy, Reddihough, & Story, 2003; Mehrholtz et al., 2005). Nevertheless, the question remains, irrespective of the scale used, whether such clinical measures are suitable to use in studies like the current study where the participants were free to move at their own pace. We therefore suggest that the relationship between proprioception measured with self-paced movement and the level of spasticity (measured with the Tardieu or the MAS) should take into account both the velocity of the self induced movement and the velocity of the passive movement used to evaluate spasticity.

Correlations between the arm/hand functionality and joint-position sense revealed that a higher QUEST score was related to a higher accuracy on the active joint-position sense task. However, this was only found for the QUEST score part A (dissociated movements) in relation with the accuracy of the impaired limb in the screen condition for the large matching distance. A possible explanation for the small amount of correlations between the QUEST and the active joint-position sense might be that the QUEST is performed under full vision. The visual information could compensate for the deteriorated joint-position sense whereas in the active joint-position sense task used in this study, no full compensation could take place since no visual feedback of the moving limb was available. Therefore, the absence of a significant relationship might indicate that on average the participants were able to compensate for the impaired proprioception with online visual control.

In sum, it can be concluded that static visual feedback of the less-impaired limb improved the active joint-position sense of the impaired limb in children with SHCP. Static mirror visual feedback did not have a detrimental effect on active joint-position sense. In addition, it was demonstrated that the distance that had to be covered by the matching limb had an influence on the differences between the limbs and the differences between the visual conditions. In general the error became smaller with a smaller matching distance. The relationship between matching accuracy and arm/hand functionality and matching accuracy and spasticity remains indecisive.

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