

The role of feedback on cognitive motor learning in children with Cerebral Palsy

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April 2017

A thesis submitted to McGill University in partial fulfillment of the requirements of the degree
of Doctorate in Neuroscience

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DEDICATION

This thesis is dedicated to my late grandfather, Laurent Théoret. You have been an inspiration through my whole life. I am eternally grateful for everything you have done for me.

I would like also to thank my parents. You have been exemplary models for me. There is no word to describe my gratitude of everything you have done for me. Merci.

Lastly, I wish my gratitude to my girlfriend Karen, for without her assistance, this PhD would have been impossible. Thank you for supporting me and for the endless discussion about science.

ABSTRACT

Cerebral Palsy (CP) is the most common pediatric neurological disorder and one of the most costly for the health system. CP impairments include muscle tone disorders (e.g., spasticity and muscle weakness), cognitive impairments and reduced sensation. Collectively, these impairments could alter reaching movements, leading to limited functional activities and limited participation levels. The use of motor learning principles to change movement behavior may increase functional activity and participation levels. Motor learning principles include practice intensity, repetitions, sensation and provision of extrinsic feedback. In particular, extrinsic feedback is defined as additional information provided to the individual. Extrinsic feedback can be provided during or after performance of a task, and delivered at different frequencies through different modalities (e.g., visual and auditory). The provision of extrinsic feedback has been suggested to improve motor learning in children with CP though there is still limited evidence in regards to which feedback modalities to prioritize due to the lack of standardized research paradigms. Thus, a more robust research design in which both the manipulation and the standardization of extrinsic feedback, such as may be done by using virtual reality systems, is needed. The global aim of this thesis was to optimize motor learning through the manipulation of extrinsic feedback in children with mild hemiplegic CP.

This thesis includes four manuscripts: three experimental studies and one literature review. The first manuscript, which is a secondary analysis of an intervention trial, examines the role of sensation on upper limb motor learning in children with hemiplegic CP. A total of 16 children with hemiplegic CP (Manual Ability Classification System Level II-IV) participated in a 15-hour intervention. The main finding was that tactile threshold and proprioception were associated with retention of improvements in velocity during a reach-to-grasp task. These findings suggest that children with poorer sensation may have a limited ability to improve movement execution resulting in sub-optimal motor learning in comparison to children who do not have reduced sensation.

Provision of extrinsic feedback is thus recommended to compensate the reduced sensation observed in children with CP in order to improve movement execution and to optimize motor learning. The second manuscript examines the role of extrinsic feedback on upper limb motor

learning in typically-developing children and in children with CP. Results of this review suggest that there is a lack of consistency in the modalities and frequencies of feedback delivery used to improve motor learning in typically-developing children and in children with CP. Moreover, since the complexity of the task influences motor learning, it has been suggested to provide continuous extrinsic feedback in early learning of a complex movement due to the higher cognitive load of faded feedback. Thus, the learner may rely more on the provision of feedback than reduced feedback to learn the desired task. Questions arising from this review indicate the importance of developing a research paradigm in which it would be possible to study and to manipulate different combinations of modalities and frequencies of extrinsic feedback to improve upper limb motor learning in typically-developing children and in children with CP.

A way to deliver an intervention to manipulate and to standardize the provision of extrinsic feedback is the use of virtual reality systems. However, the object's perceived location in a virtual environment may be altered, affecting movement execution. Thus, the objective of the third manuscript was to compare reaching kinematics made in three planes (sagittal, frontal, longitudinal) in a low-cost, game-based virtual reality system to those made in a matched physical environment in typically-developing children and children with mild hemiplegic CP. The main finding was that all reaching gestures made in the virtual environment in comparison to those made in a physical environment were slower and required less trunk flexion and rotation in typically-developing children. In children with CP, trajectories were more curved and less trunk flexion and rotation were used for each movements made in the virtual environment. Lastly, kinematics of all three gestures did not differ between typically-developing children and children with CP suggesting that they tend to interact using similar movement patterns. These results demonstrate that a low-cost, game-based virtual reality system could be used as an adjunct therapy to improve reaching in children with mild hemiplegic CP when taking into account the differences in kinematics.

The fourth manuscript determines the feasibility of delivering different frequencies of extrinsic feedback through a virtual reality system and investigates their possible effects on improving upper limb motor learning in children with mild hemiplegic CP. A total of 7 children with mild hemiplegic CP participated in this 7-day intervention study. The results showed that all children

were able to complete all clinical and experimental sessions. Motivation was considered high in all children. Hence, implementation and acceptability feasibility criteria of this study were considered satisfied. The findings also showed that all clinical scores were maintained or improved. Kinematic changes were also observed in some children. This pilot study demonstrates the possibility of manipulating and standardizing extrinsic feedback in children with mild hemiplegic CP with the objective of improving upper limb motor learning.

The results of this thesis highlight the importance of using principles of motor learning, such as extrinsic feedback, to improve movement execution and to optimize motor learning in children with mild hemiplegic CP. The use of virtual reality systems is proposed to develop a robust research paradigm as it offers the possibility to of manipulating and standardizing extrinsic feedback.

RÉSUMÉ

La Paralyse Cérébrale (PC) est un ensemble de troubles neurologiques des plus fréquents en pédiatrie et un des plus coûteux pour le système de la santé. Les déficits de la PC incluent des problèmes de tonus musculaires (ex. spasticité et faiblesse musculaire), des déficits cognitifs et une réduction de la sensation. Ces déficits ont comme impact d'altérer les mouvements d'atteintes diminuant ainsi la capacité fonctionnelle et ultimement, limite la participation sociale.

L'utilisation des principes d'apprentissage moteur dans le but de modifier l'exécution du mouvement peut améliorer les capacités fonctionnelles et le niveau de participation. Ces principes d'apprentissage moteur incluent l'intensité de la pratique, la répétition, la sensation et la provision de feedback extrinsèque. En particulier, le feedback extrinsèque est défini comme une information additionnelle fournie à l'individu qui peut être administrée pendant ou après la tâche, et ce, à diverses fréquences sous diverses modalités (ex. visuelle et auditive). Malgré le manque d'évidence scientifique et une absence de standardisation dans les protocoles de recherche pour la modalité qui devrait être priorisée pour optimiser l'apprentissage moteur, l'utilisation du feedback extrinsèque est suggérée pour améliorer l'apprentissage moteur chez les enfants avec la PC. Ainsi, un projet de recherche robuste dans lequel la manipulation et la standardisation du feedback extrinsèque est possible, tel que l'utilisation possible de systèmes de réalité virtuelle, est nécessaire. Le principal objectif de cette thèse est d'optimiser l'apprentissage moteur grâce à la manipulation du feedback extrinsèque chez les enfants avec la PC de type hémiplégique légère.

Cette thèse inclut 4 manuscrits: trois études expérimentales et un article de revue. Le premier article scientifique, qui consiste à une analyse secondaire d'une étude déjà publiée, évalue le rôle de la capacité sensorielle pour l'apprentissage moteur du membre supérieur chez les enfants avec une PC de type hémiplégique. Un total de 16 enfants avec la PC de type hémiplégique (Manual Ability Classification System Level II-IV) ont participé à une intervention d'une durée de 15 heures. Les résultats de cette étude démontrent une association entre le seuil tactile et la proprioception avec le maintien de l'amélioration de la vitesse durant une tâche d'atteinte. Ces résultats démontrent que les enfants qui ont une diminution de sensation peuvent être limité dans leur exécution motrice, ce qui a comme impact de sous-optimiser l'apprentissage moteur en comparaison avec des enfants qui n'ont pas une diminution de la sensation.

Ainsi, afin de compenser pour la diminution de sensation observée chez les enfants avec la PC, l'utilisation du feedback extrinsèque est recommandée afin d'améliorer l'exécution motrice et pour optimiser l'apprentissage moteur. Le deuxième manuscrit évalue le rôle du feedback extrinsèque sur l'apprentissage moteur du membre supérieur chez les enfants avec un développement typique et ceux avec la PC. Les résultats de cette revue scientifique suggèrent un manque de cohérence dans l'utilisation des modalités et des fréquences de feedback extrinsèque pour permettre l'amélioration de l'apprentissage moteur chez les enfants avec un développement typique et ceux avec la PC. De plus, puisque la complexité de la tâche peut avoir un impact sur l'apprentissage moteur, il est suggéré d'offrir du feedback en fréquence continue au début de l'apprentissage d'une tâche complexe. Ainsi, la personne pourra davantage dépendre sur le feedback continu en comparaison du feedback réduit pour apprendre la tâche. La raison évoquée est que le feedback en fréquence continue ne nécessite pas une grande charge cognitive. Plusieurs questions ont été soulevées suite à cette revue. Par exemple, il est important de développer un projet de recherche permettant d'étudier et de manipuler différentes combinaisons des modalités et des fréquences de feedback extrinsèque avec l'objectif ultime d'améliorer l'apprentissage moteur des membres supérieurs chez les enfants avec un développement typique et ceux avec la PC de type hémiplégique.

L'utilisation des systèmes de réalité virtuelle a comme avantage de pouvoir manipuler et standardiser le feedback extrinsèque offert durant une intervention. Cependant, la perception des objets dans un environnement virtuel peut être modifiée, ce qui peut avoir un impact négatif sur l'exécution du mouvement. Ainsi, le troisième manuscrit a comme objectif de comparer des mouvements d'atteintes effectués dans trois plans (frontal, sagittal et longitudinal) dans un système de réalité virtuelle de type jeu vidéo à faible coût avec les mouvements effectués dans un environnement physique chez les enfants avec un développement typique et ceux avec une légère PC de type hémiplégique. Le résultat principal est que tous les mouvements d'atteintes effectués dans un environnement virtuel en comparaison à ceux effectués dans un environnement physique sont plus lents et nécessitent moins de flexion et rotation du tronc chez les enfants avec un développement typique. Pour ce qui est des enfants avec la PC, les trajectoires de chacun des mouvements effectués dans un environnement virtuel sont plus courbées et l'utilisation de la

flexion et rotation du tronc est moins proéminente. Finalement, la cinématique des mouvements effectués dans les trois plans par les enfants avec un développement typique et ceux avec la PC ont des similarités suggérant l'utilisation de patron moteur similaire. Ces résultats démontrent qu'un système de réalité virtuel de type jeu vidéo à faible coût peut être utilisé comme une thérapie additionnelle pour améliorer les gestes d'atteintes chez les enfants avec la PC de type hémiplégique. Cependant, il est important de prendre en considération les différences cinématiques observées dans cette étude.

L'objectif du quatrième manuscrit est de déterminer la faisabilité de transmettre différentes fréquences de feedback extrinsèque à partir d'un système de réalité virtuelle et d'évaluer l'impact sur l'amélioration de l'apprentissage moteur au niveau des membres supérieurs chez les enfants avec une légère PC de type hémiplégique. Un total de sept enfants avec la PC de type hémiplégique ont participé à une étude d'une durée de 7 jours (4 jours d'intervention et 3 jours d'évaluation). Les résultats démontrent que tous les enfants ont été en mesure de compléter les mesures cliniques et cinématiques. Tous les enfants ont mentionné une motivation élevée durant l'intervention. Ainsi, les critères d'implémentation et de réussite de faisabilité de cette étude sont considérées positif. Les résultats ont aussi démontrés une amélioration de la performance dans les échelles cliniques. Des changements significatifs des mesures cinématiques ont aussi été observés chez tous les enfants. Cette étude pilote démontre la possibilité de manipuler et de standardiser le feedback extrinsèque chez les enfants avec la PC de type hémiplégique tout en ayant l'objectif de favoriser l'apprentissage moteur des membres supérieurs.

Les résultats de cette thèse mettent l'emphasis sur l'importance d'utiliser les principes d'apprentissage moteur tels que le feedback extrinsèque pour améliorer l'exécution du mouvement et pour optimiser l'apprentissage moteur chez les enfants avec une PC de type hémiplégique. L'utilisation des systèmes de réalité virtuelle est proposée pour développer un paradigme de recherche robuste dans lequel il est possible de manipuler et de standardiser le feedback extrinsèque.

ACKNOWLEDGEMENTS

I would like to thank my thesis supervisor, Dr. Mindy F. Levin, for her constant mentoring over the last five years. This thesis would not have been possible without her help and guidance. Her valuable knowledge on science and her positive criticism helped me to be a better researcher.

I wish to thank Dr. Anatol Feldman for always being supportive and being open to discuss about my ideas.

I would also like to thank my committee members, Dr. Laurie Snider and Dr. Martin Lemay for their support during the last five years.

Thank you to Karen Fung for her proof reading and the amazing support throughout the PhD. I am eternally grateful for the help.

Special thanks to all my colleagues at the Sensorimotor Control and Rehabilitation Laboratory of the Jewish Rehabilitation Hospital for all their supports. I would also like to acknowledge Valeri Goussev for the amazing program he has made.

The project would not have been feasible without the salary support of the Vanier Canada Graduate Scholarships (Canadian Institutes of Health Research) and the Fonds de recherche du Québec - Santé (FRQS). In addition, I wish to acknowledge the Réseau Provincial de recherche en Adaptation-Réadaptation (REPAR) for their funding support.

PREFACE AND CONTRIBUTION OF AUTHORS

Thesis format

This thesis is manuscript-based, and is prepared according to the McGill Graduate and Postdoctoral Studies guidelines for thesis preparation. This thesis contains four original papers, of which two are published, one is submitted to a peer-reviewed journal and one that will be submitted to a peer-reviewed journal.

Chapter 1

Chapter 1 outlines the rationale and provides a short introduction for this thesis and the objectives.

Chapter 2

Chapter 2 is a literature review composed of 6 sections. The first section describes the etiology and the classification of Cerebral Palsy. The second section briefly describes how movements are made and classified. The third section defines motor learning as well as certain theories on how an individual learns a task. Different principles that could optimize motor learning and improve movement execution are then described. The fourth section explores the multisensory integration theory. The fifth section discusses the use of virtual reality systems in rehabilitation as well as its advantages and possible limitations. The last section presents the rationale of this thesis.

Chapter 3

Chapter 3 features the first manuscript, which is a secondary analysis of an intervention trial that examined the role of sensation on upper limb motor learning in children with spastic hemiplegic Cerebral Palsy.

Chapter 4

Chapter 4 consists of the second manuscript which is a review that identified the most effective modalities and frequencies of feedback for improving upper limb motor skills in typically-developing children and children with Cerebral Palsy.

Chapter 5

Chapter 5 contains the third manuscript in which reaching kinematics made in different planes in a virtual reality system were compared to those made in a matched physical environment in typically-developing children and in children with Cerebral Palsy.

Chapter 6

Chapter 6 features the fourth manuscript, which addressed the feasibility of manipulating extrinsic feedback in a virtual reality system in children with mild hemiplegic Cerebral Palsy.

Chapter 7

Chapter 7 discusses the results from all four manuscripts in regards to the content presented in the literature review.

Manuscript # 1 (Chapter 3) - The data analysis and the preparation of the manuscript was done by Maxime Robert under the supervision of Dr. Mindy Levin. The project was designed by Dr. Mindy Levin and Dr. Heidi Sveistrup. Sheila Schneiberg performed the recruitment and the data collection under the supervision of Dr. Mindy Levin. Gevorg Chilingaryan provided statistical guidance. All authors read and approved the final version of the manuscript submitted for publication.

Manuscript # 2 (Chapter 4) - The extensive review of the literature, the methodology design, the scoring of each individual studies and the preparation of the manuscript were completed by Maxime Robert and Krithika Sambasivan under the supervision of Dr. Mindy Levin. Dr. Mindy Levin resolved conflicts with the scoring of the articles' quality assessment. All authors read and approved the final version of the manuscript submitted for publication.

Manuscript # 3 (Chapter 5) – Under the supervision of Dr. Mindy Levin, the study design, the kinematic data collection, the data analysis, the statistical analysis and the manuscript preparation were performed by Maxime Robert. Dr. Mindy Levin advised in all phases of the project as well as critically edited the manuscript. All authors read and approved the final version of the manuscript submitted for publication.

Manuscript # 4 (Chapter 6) – The study design, the kinematic data collection, the data analysis, the statistical analysis and the manuscript preparation were completed by Maxime Robert under the supervision of Dr. Mindy Levin. Dr. Laurie Snider and Dr. Martin Lemay contributed constructive criticism about the study design. The clinical data was collected by Daniela Chan-Viquez and Marika Demers. Dr. Mindy Levin critically edited the manuscript. All authors read and approved the final version of the manuscript submitted for publication.

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LIST OF ABBREVIATIONS

CP: Cerebral Palsy

CRIR: Centre for Interdisciplinary Research in Rehabilitation

IC: Index of Curvature

ICFDH: International Classification of Functioning, Disability and Health

IMI: Intrinsic Motivation Inventory

IREDS: Infrared-Emitting Diodes

KP: Knowledge of Performance

KR: Knowledge of Results

MACS: Manual Ability Classification System

NIH: National Institute for Health Research

NTR: No Trunk Restraint

PC: Paralysie Cérébrale

PE : Physical Environment

PVL: Periventricular Leukomalacia

PWMI : Periventricular White Matter Injury

RCT: Randomized Control Trials

ROM : Range of Motion

S-W: Semmes-Weinstein

TD: Typically-Developing

VE: Virtual Environment

VR: Virtual Reality

WTR: With Trunk Restraint

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CHAPTER 1 - GENERAL INTRODUCTION

1.1 Background

Cerebral palsy (CP) is one of the most common childhood disabilities with a prevalence of 1.5 to 2.5 per 1000 births (Paneth et al., 2006). Children with CP have impairments leading to altered movements, which may decrease their functional activities and their participation compared to their typically-developing (TD) peers (Klingels et al., 2012; Sakzewski et al., 2009). More specifically, children with CP may present muscle tone disorders, reduced sensation, muscle weakness, decreased range of motion, deficits in interjoint coordination, and/or deficits in motor planning, all of which may lead to altered movement behavior. These changes in movement behaviour may contribute to difficulties in performing actions, such as reaching tasks (Eliasson and Gordon, 2000). For instance, children with hemiplegia have prolonged movement times and increased use of trunk flexion when reaching for targets located within maximal arm's length compared to TD children. Interventions aim to reduce the difficulties experienced during performance of reaching tasks by improving movement behaviour. One way to improve movement behaviour is through the manipulation of principles of motor learning, such as the number of task repetitions, the level of task difficulty, extrinsic feedback and motivation (Kleim and Jones, 2008; Molier et al., 2010; Schmidt and Lee, 2011). Feedback is crucial to deliver information on the quality of the movement and of the performance. Feedback on parameters of a movement can be provided through different sensory modalities (i.e., visual or auditory) and at different frequencies (i.e., continuous or faded; (Sigrist et al., 2013)). When given additional feedback, healthy adults, individuals with stroke, TD children and children with CP demonstrated better retention of new motor skills in comparison to their peers who did not receive any feedback (Abadi et al., 2014; Hemayattalab and Rostami, 2010; Talbot and Junkala, 1981). However, there are contradictory findings on which modalities and frequencies of extrinsic feedback should be prioritized to optimize upper limb motor learning. As an adjunct to conventional therapy, virtual reality (VR) systems offer the possibility of customizing and standardizing feedback provided to children, in order to improve motor learning during a training intervention. To date, research on feedback provision and on development of VR technology is primarily focused on healthy adults and on individuals with stroke. There is yet any research paradigm that investigates the optimal feedback frequency for motor learning in children with

CP. Thus, it is imperative to develop and to test the feasibility of delivering and of standardizing different modalities of extrinsic feedback using a VR system with the objective to improve movement execution and motor learning in children with CP.

1.2 Objectives of the Thesis

To optimize upper limb motor learning through the manipulation of extrinsic feedback in children with hemiplegic CP

The specific objectives of this thesis are:

- 1) To determine if upper limb kinematics in children with CP during a standardized reach-to-grasp task could be learned, retained and transferred to a similar task;
- 2) To characterize the role of sensation on motor learning in children with hemiplegic CP;
- 3) To identify knowledge and gaps on the use of extrinsic feedback to improve upper limb motor skills in TD children and in children with CP;
- 4) To compare upper limb and trunk kinematics of reaching made in two planes in a 2D virtual environment to those made in a physical environment in TD children and children with CP
- 5) To determine the relationships between sensory impairments and reaching kinematics in children with mild hemiplegic CP;
- 6) To determine the feasibility (i.e., implementation and acceptability) of delivering extrinsic feedback through a VR system for upper limb skill acquisition in children with mild hemiplegic CP;
- 7) To determine the feasibility of implementing a short intensive VR intervention in children with mild hemiplegic CP.

CHAPTER 2 - LITERATURE REVIEW

2.1 Cerebral Palsy

First defined by William Little in 1843, CP was referred to as Little's disease (Johnston and Hoon, 2006; Jones et al., 2007). Little described CP as a disorder that affects motor development in the first years of life. Since then, a number of definitions have been proposed. Until recently, there has been no general consensus among clinicians and researchers on the definition of CP. Rosenbaum et al. (2005) proposed the following definition: "*Cerebral palsy (CP) describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, cognition, communication, and behaviour, by epilepsy, and by secondary musculoskeletal problems*". The purpose of this new definition is to demonstrate the multidimensionality of impairments in children with CP. This definition facilitates the diagnosis and classification (see below) of CP as well as assessments of functional abilities. Although Rosenbaum et al. (2007) emphasized the non-progressive nature of CP in their description, secondary conditions can occur increasingly with age due to the sedentary lifestyle of children with CP, which perpetuates a cycle of degeneration of their health condition (Verschuren et al., 2012).

2.1.1 Prevalence and Incidence

Depending on the country, the prevalence of CP generally ranges from 1.5 to 2.5 per 1000 live births and has been relatively stable throughout the world over the past 40 years (Kuban and Leviton, 1994; Paneth, 1986; Paneth et al., 2006). In Quebec, Canada, the incidence of CP is 2.09 in every 1000 births (Self et al., 2010). A modest increase was observed in the 1980s (Paneth et al., 2006), but the prevalence has since decreased over the last decade as explained by better care for low birth weight infants. Due to its high incidence and its high cost for the health care system (Johnston and Hoon, 2006), CP is one of the most studied types of neurodevelopmental disorders in children (Bishop, 2010)

2.1.2 Etiology

The causes of CP can be explained by a large number of intrinsic and extrinsic factors. There are also interpersonal differences for the causes of CP. Understanding the etiology of CP enhances information that will ultimately be used to optimize clinical care (Miller, 2007). The broad etiology of CP varies from a malformation of the infant's brain to consequences of excessive drug or alcohol use during pregnancy. In 1897, Freud suggested that CP was caused by a delay in brain development before birth (Johnston and Hoon, 2006). In 1956, Minear proposed that an injury to the developing brain at prenatal, perinatal or postnatal time points can lead to CP. Consequently, Miller (2007) suggested that the etiology should be classified by when the brain lesion occurs: prenatal, perinatal, postnatal periods.

Firstly, prenatal risk factors affecting brain development have been identified in 70 to 80% of term infants who develop CP (Johnston and Hoon, 2006). Among those risk factors, 10 to 15% of the prenatal cases arise from intrapartum complications such as hypoxia, contagious disease or traumas (Bialik and Givon, 2009; Johnston and Hoon, 2006). On the other hand, spastic diplegic CP in children without evidence of brain abnormalities is hypothetically caused by genetic factors (Numata et al., 2013). Epidemiological studies also show that maternal thyroid dysfunction is a major cause of CP (LaFranchi et al., 2005). Other factors underlying the different types of CP during the prenatal period include infection, excess of drugs during pregnancy, excess of alcohol, maternal epilepsy, mental retardation of the mother or bleeding during the third trimester of pregnancy (Bialik and Givon, 2009).

In the perinatal period, low birth weight and prematurity are two of the most common risk factors for CP (Bialik and Givon, 2009; Msall, 2004). Premature birth (i.e., before 37 weeks of gestation) explains 25 to 40% of the incidence of CP (Johnston and Hoon, 2006). Of premature births, 10 to 15% of newborns with a birth weight under 1500 grams are at risk of developing CP (Bialik and Givon, 2009; Johnston and Hoon, 2006; Jones et al., 2007; Sankar and Mundkur, 2005). Neuroimaging studies of infants demonstrated that the risk of developing CP was related to intraventricular hemorrhage. Since decades ago, intraventricular hemorrhage was a known risk for premature infants as the intracerebral blood vessels were described as extremely fragile

during this developmental period, leading to wide oscillations in cerebral blood flow. Presently, the incidence of intraventricular hemorrhage has dramatically decreased, possibly explained by advances in mechanical ventilation and improvement of care enabling the reduction of fluctuations in cerebral blood flow (Kent et al., 2012). One of the most important risk factors of CP nowadays is injury to deep white matter of the premature brain (Bialik and Givon, 2009; Johnston and Hoon, 2006). The development of the white matter adjacent to the ventricle is highly vulnerable prior to 32 weeks of gestation, which explains why preterm children are more at risk of developing CP (Johnston and Hoon, 2006). In fact, periventricular white matter injury (PWMI) has been specifically recognized as the most common brain abnormality in preterm children with CP (Johnston and Hoon, 2006). PWMI occurs due to various factors including vulnerability of oligodendrocytes before 32 weeks of gestation (Johnston and Hoon, 2006), ischemia and infection (Back et al., 2005). The most prevalent cause of PWMI is periventricular leukomalacia (PVL). PVL is defined as an injury to the deep white matter of the premature brain due to cystic necrotic lesions (Back et al., 2005; Johnston and Hoon, 2006). Only 12% of children born at term with spastic diplegic CP have PVL, reinforcing the idea that PVL is mostly associated with preterm children (Koeda et al., 1990; Melhem et al., 2000). PVL typically results in thinning of the posterior body of the corpus callosum, enlargement of the lateral ventricles and irregularity of the lateral ventricular walls (Melhem et al., 2000). Furthermore, PVL is associated with cognitive and sensory abnormalities (Marlow, 2004). Melhem et al. (2000) demonstrated a direct relationship between the magnitude of white matter injury and the severity of motor and cognitive impairments.

Thus, most cases of hemiplegic CP are associated with either middle cerebral artery infarction, congenital cortical-subcortical lesions or PWMI (Feys et al., 2010; Wimalasundera and Stevenson, 2016). Of all the cases of CP, the incidence of CP from the postnatal period is 12 to 21% (Johnston and Hoon, 2006). For this smaller proportion of CP cases, there exist a higher number of risk factors including, but not limited to head trauma, meningitis, encephalitis, toxicities, shaken baby syndrome and brain infarcts (Bialik and Givon, 2009; Johnston and Hoon, 2006; Sankar and Mundkur, 2005).

In summary, the etiology of CP differs from child to child, but the causes can be categorized by the timing of their occurrence. Prenatal risk factors have been identified in 10% of children who develop CP, whereas 21 to 40% of CP causes arise in the perinatal period and 12 to 21% of the cases are attributed to postnatal factors (Sankar and Mundkur, 2005). Today, the cause of approximately 30% of CP cases remains idiopathic (Jones et al., 2007; Rosenbaum et al., 2007).

2.1.3 Classification

The classification of CP has evolved over the last decades due to the enhanced diagnoses of this neurodevelopmental disorder in children. Variations in CP classification exist because of the heterogeneity in sensorimotor impairments possible with CP. The classification of CP provides a clinically useful overview of the different sensorimotor impairments. In addition, a variety of classification systems are still used today in order to give a description as pertinent as possible to the CP case studied. The basic classification of CP types is generally based on the anatomic distribution of motor impairment (i.e., hemiplegic, diplegic, quadraplegic) and muscle tone disorders (e.g., spastic, ataxia, etc.). Building upon the basic classification, a latter one provides additional information on the causes or the risk factors of CP. However, this oversimplified classification tends to exclude the other potential impairments and their impact on the health of children with CP. The next paragraph presents a classification based on anatomic distribution as a broad overview. Details on the classification of muscle tone disorders, which is usually provided in reports relating CP are discussed in the section on all sensorimotor impairments.

2.1.3.1 Classification Based on Anatomical Distributions

The anatomical distribution of the impairments classify CP into 5 types: diplegia, hemiplegia, quadriplegia, and the less prevalent forms of monoplegia and triplegia (Minear, 1956). The term “diplegia” refers to impairment of both limbs on either the lower or the upper body. Diplegia is more commonly observed in the lower limbs than in the upper limbs (Bialik and Givon, 2009). “Hemiplegia” is defined as impairment of one side of the body (Rosenbaum, 2007). “Quadriplegia” refers to all four limbs being affected (Bialik and Givon, 2009). The terms

“monoplegia” and “triplegia” respectively designate impairment in one and three limbs (Bialik and Givon, 2009). In CP, diplegia is the most common form (30-40%) followed by hemiplegia (20-30%) and quadriplegia (10-15%; (Sankar and Mundkur, 2005).

2.1.3.2 International Classification of Functioning and Disability and Health

In 2001, the World Health Organization (WHO) published the International Classification of Functioning, Disability and Health (ICF; WHO, 2001). The objective of the ICF is to provide a scientific basis to understand and to study health and health-related states, outcomes and determinants for any population. In sum, the framework of the ICF provides information in a meaningful, interrelated and easily accessible way. The next section describes the different classifications within the ICF. The ICF has been used to describe the details of different CP types; the ICF does not classify individuals, but rather it describes a situation within an array of health and health-related domains (WHO, 2001). The ICF categorizes health and health-related domains into two components: 1) functioning and disability, and 2) contextual factors. The ICF model can be applied to healthy individuals as well as to people living with a disability. The functioning and disability section is divided into three parts: 1) Body Functions and Structures, 2) Activity And 3) Participation at the societal level. The second section of the ICF identifies contextual factors including environmental and personal barriers to health. However, these barriers are not discussed as they are beyond the objective of this thesis. The following section explores the different impairments and their impact on the Activity and Participation levels based on the components of the ICF.

2.1.3.3 Body Functions and Structure Domain

Generally, muscle tone disorders in CP can be classified in two categories: the spastic form and the non-spastic form (dyskinesia/ataxia;(Himpens et al., 2008). Generally used to identify the subtypes of CP, this classification does not categorize other health domains (e.g., sensory impairments, muscle weakness, cognitive deficits, etc.) as done in the ICF model. Thus, the use of the ICF model to describe all health domains of the Body Function and Structure Level will be

discussed in this section. Within the Body Function and Structure Level, the musculoskeletal system is the most studied impaired system in children with CP in comparison to cognitive and sensory systems due to its high prevalence. The diversity in the muscle tone disorders in CP is not surprising as it reflects the various etiological factors possible during each period of gestation as discussed in Section 2.1.2 (Feys et al., 2010). A multitude of studies aimed to relate the lesions to the severity of neurological impairments, but their efforts remain non-conclusive (Staudt et al., 2004; Truwit et al., 1992; Wiklund and Uvebrant, 1991). Among the different muscle tone disorders, spasticity is the most common type of hypertonicity, which is present for 70% of children with CP (Rosenbaum et al., 2007; Sanger, 2005; Taft, 1995). Spasticity is defined as a motor disorder characterized by a velocity-dependent increase in tonic stretch reflexes (muscle tone) with exaggerated tendon jerks, resulting from hyperexcitability of the stretch reflex, as one component of the upper motor neuron syndrome (Johnson, 2002; Lance, 1980). Although the pathophysiological mechanisms of spasticity remain unclear, general consensus recognizes the roles of damage to the corticospinal and other descending pathways (Jones et al., 2007; Young, 1994) in addition to changes in muscle properties, such as muscle fiber type and mechanical properties, and associations of etiology and lesion location (Nielsen et al., 1995; Sanger et al., 2003). More importantly, the pathology of spasticity in children is affected by the reorganization of the supraspinal input and by the ongoing motor maturation (Bar-On et al., 2015). Spasticity has the effect of a strong facilitation of synaptic transmission in the 1a sensory fibers in the monosynaptic reflex pathway (Kandel et al., 2000). In addition, several studies suggested that Type II afferent fibers from the muscle spindles activate the alpha motoneurons in spasticity (Trompetto et al., 2014). The most severe form of spasticity often arises from a brain maldevelopment occurring in the prenatal stage (Wimalasundera and Stevenson, 2016).

The other 20 to 30% of CP cases may have different types of hypertonic disorders such as dyskinesia, ataxia or a mixed form (Bialik and Givon, 2009; Himpens et al., 2008; McManus et al., 2006). “Dyskinesia” is defined as involuntary, uncontrolled, recurring and occasionally stereotypical movements (Rosenbaum et al., 2007). Most of the time, injuries of the basal ganglia occurring around the time of birth result in bilateral and dyskinetic types (Rosenbaum et al., 2007). However, the severity of the damage can increase with prolonged duration of the insult.

Dyskinesia is classified by subgroups of motor disorders of varying degrees from dystonic to chorea-athetotic forms. “Dystonia” refers to an abnormal posture, while “chorea” designates rapid involuntary movement and “athetosis” signifies slower movements (Johnston and Hoon, 2006). However, prolonged hypoxia may result in a mixed motor pattern and the appearance of additional comorbidities. With a prevalence of around 5% (Shepherd, 1994), the ataxic form of CP is defined as a loss of orderly muscular coordination, resulting in an abnormal force, rhythm and accuracy during the performance of functional tasks (Rosenbaum et al., 2007). Finally, the uncommon mixed form of CP accounts for less than 2.5% of children with CP (Stanley et al., 2000). One school of thought suggested that the term “mixed form” should be avoided, and that only the most dominant clinical form should be named in order to avoid confusion (Rosenbaum et al., 2007). Muscle tone disorders, particularly spasticity, were once questioned as to whether or not they have any impact on functional outcomes of the upper limbs (Kim and Park, 2011). Recent findings correlated the severity of upper limb muscle tone disorders with activity levels based on the scoring of Melbourne Assessment and the Assisting Hand Assessment clinical scales (Klingels et al., 2012). A later study found that muscle tone in the wrist was highly correlated with the aforementioned assessments. Therefore, the severity of spasticity and other muscle tone disorders need to be taken into consideration when measuring the level of activity in children with CP. Furthermore, a review suggested that Botulinum Toxin injections in the flexor and/or extensor elbow muscles reduce spasticity in children with CP, leading to increased functional activity as measured by the Melbourne Assessment, the Jebsen-Taylor Test, the Assisting Hand Assessment or the Quality of Upper Extremity Skills Test (Sakzewski et al., 2009). In this meta-analysis, effect sizes and mean differences were used to measure the improvements of individual studies reported. Overall, the effect of Botulinum Toxin injections was found to be small to moderate when added to an intensive therapy such as constraint-induced movement therapy and hand-arm bimanual intensive training when compared to the control group who did not receive the injection.

In addition to muscle tone disorders, a review by Mockford et al. (2010) showed that every child with CP also has some degree of muscle weakness (Damiano et al., 2002; Shortland, 2009). In the study of muscle strength in 60 children with spastic CP, Ross et al. (2002) found that all participating children with spastic CP were significantly weaker in comparison to TD peers. The

causes of muscle weakness are proposed to be related to decreased activation in descending pathways (Mockford and Caulton, 2010), lack of physical activities (Fowler et al., 2007), changes in muscle volume (Barrett and Lichtwark, 2010), and spasticity (Engsberg et al., 2000). Although previously, muscle weakness was not considered as a predictor of manual ability in children with CP, this notion has since been challenged (Arnould et al., 2007; Sakzewski et al., 2009; van Meeteren et al., 2007). For instance, handgrip strength is correlated with both unimanual and bimanual activities (Braendvik et al., 2010). Other factors of muscle weakness in CP is the shortening of muscle fibers, particularly in comparison to age-matched TD children (Boyd and Winstein, 2001; Shortland, 2009). These factors not only increase muscle weakness, but may lead to a reduction in the range of motion of joints in both lower and upper limbs (Mutlu et al., 2007).

In addition to muscular impairments, some children with CP have reduced sensation for light touch, tactile threshold, level of pain and proprioception (Bax et al., 2005; Krigger, 2006). Reduced sensation in children leads to difficulty in refined hand functions, such as grip force and tactile exploration (Clayton et al., 2003; Majnemer et al., 2010). In cases of the most severe sensory deficits, children with CP neglected the affected limb resulting in the deterioration of the affected arm and thus, in altered movement execution (McLaughlin et al., 2005; Thibault et al., 1994). Reduced sensation is also linked to limitations in activity level (Arnould et al., 2007; Klingels et al., 2012; Sakzewski et al., 2009).

More than 2/3 of children with CP have cognitive deficits or learning disorders (Jones et al., 2007). These cognitive deficits need to be considered as they could negatively affect motor planning resulting in altered movement execution (Steenbergen and Gordon, 2006). Children with CP may also have visual impairments, such that 75% of children with spastic CP have strabismus (Taft, (1995). Visual impairments could lead to misinterpretation of the location of an object in the environment, which is further discussed in Section 2.7.3.

To summarize, the impairments described above may alter movement execution, which in turn limits the activity level and reduces participation in children with CP. In the next section, the impact of these impairments on reaching behaviour is discussed (Figure 2-1).

2.1.3.4 Activity Level Domain

The activity level in children with CP is different in comparison to that of their healthy peers, partially because of the impairments discussed in Section 2.1.3.3 and altered movement behaviour. Based on the ICF model (Activity Level), this section elaborates on how upper limb movements are altered and how activity levels are reduced.

The proposed standard classification of the Manual Ability Classification System (MACS) describes the impact of upper limb impairments on the child's ability to perform daily activities. The MACS is a five-level ordinal scale classifying the ability of children to manipulate objects in daily activities (Chin et al., 2005; Eliasson et al., 2006; McConnell et al., 2011). A score of MACS level V characterizes a child's ability to handle objects easily and successfully, but with slight limitation on tasks requiring speed and accuracy (Morris et al., 2006). The MACS scale proposes that no difference can be observed between the age of 4 and 18 years old (Ohrvall et al., 2014).

Deficits in coordination in children with CP can increase the difficulty of grasping and releasing objects, interfering with their ability to independently accomplish activities of daily living (Eliasson and Gordon, 2000). Comparing reaching and grasping of two different sized objects (small and large diameters) in children with CP, a study found that movement time is longer when reaching for small objects (Coluccini et al., 2007). The increase of movement time has been associated with the releasing phase of the reach-to-grasp task (Coluccini et al., 2007). The reach-to-grasp task also requires greater use of trunk rotation and flexion in children with CP in comparison to the same task performed by TD children (Ju et al., 2010).

Reaching tasks by children with CP were reported to be less precise in comparison to that of their TD peers (Sanger, 2006). Children with hemiplegia aged between 10 and 17 years old also displayed slower movement compared to TD children due to decreased shoulder flexion and elbow extension. Based on a systematic review of reaching in children with CP, the tasks took longer time to complete, but movements by children with CP had higher peak velocities in

comparison to TD children, indicating less graded control (Visicato et al., 2014). This review also reported that the endpoint trajectory path was more curved in children with CP when compared to TD children during a reaching task (Visicato et al., 2014). Essentially, the severity of the children's impairments and the complexity of the task influence the performance.

For most activities of daily living, including brushing teeth, making a sandwich and dressing, children with hemiplegic CP often have similar levels of difficulty in the completion of these tasks in comparison to TD children (Van Zelst et al., 2006). While children with mild CP can do these tasks independently, children with moderate to severe CP often require either more time or assistance from others to accomplish the same activities (Van Zelst et al., 2006; Voorman et al., 2006).

Based on the conclusion of a systematic review, activity levels in children with mild hemiplegic CP slightly interfere with the quality of life as measured in the studies reviewed (Visicato et al., 2014). In more severe cases of CP, the activity levels are greatly reduced, which moderately interferes with the quality of life. The large range of sensorimotor impairments and the reduced activity levels generally limit the participation level in children with CP (Chen et al., 2013; Chen et al., 2014; Klingels et al., 2012).

2.1.3.5 Participation Level Domain

Participation levels in children with CP is disturbingly reduced in comparison to their healthy peers, but relatively little is known about the reasons for this difference (Fernhall and Unnithan, 2002). For example, measuring the activity level of children with CP through an activity monitor found that they participate in significantly fewer sport activities, approximately a quarter to half that of their healthy peers (Bjornson et al., 2007). Furthermore, the overall amount of physical activities in which children engage in, is strongly associated with the severity of sensorimotor impairments (Maher et al., 2007).

As for the participation level at school, children with mild CP with normal cognitive development are usually integrated in mainstream school education. However, children with CP

who have severe learning disabilities and a low intelligent quotient often attend special schools (Beckung and Hagberg, 2002).

As for the social participation of children with CP, their parents reported a decreased level in comparison to their healthy siblings (Arnaud et al., 2008). However, the authors emphasized that the stress level of the parents have to be taken into consideration (Arnaud et al., 2008).

Similarly, the participation level in leisure activities of children with CP was reported to be lower in community-based activities in comparison to healthy peers (Majnemer et al., 2008).

Factors that explain why children with CP struggle to participate in sports with TD children may be explained by the demands for children with CP to be at an equivalent skills level (Verschuren et al., 2012). In the community, sport facilities may not be accessible for children with disabilities. Furthermore, facilities promoting the participation of children with disabilities often have a waiting list (Shimmell et al., 2013). Financial restrictions can also be a social barrier for families since a child with a physical disability requires costly specific care and/or adjustments (Stewart et al., 2012). Thus, participation levels in children with CP appears to affect their quality of life and may be indirectly linked with the severity of sensorimotor impairments (Klingels et al., 2012).

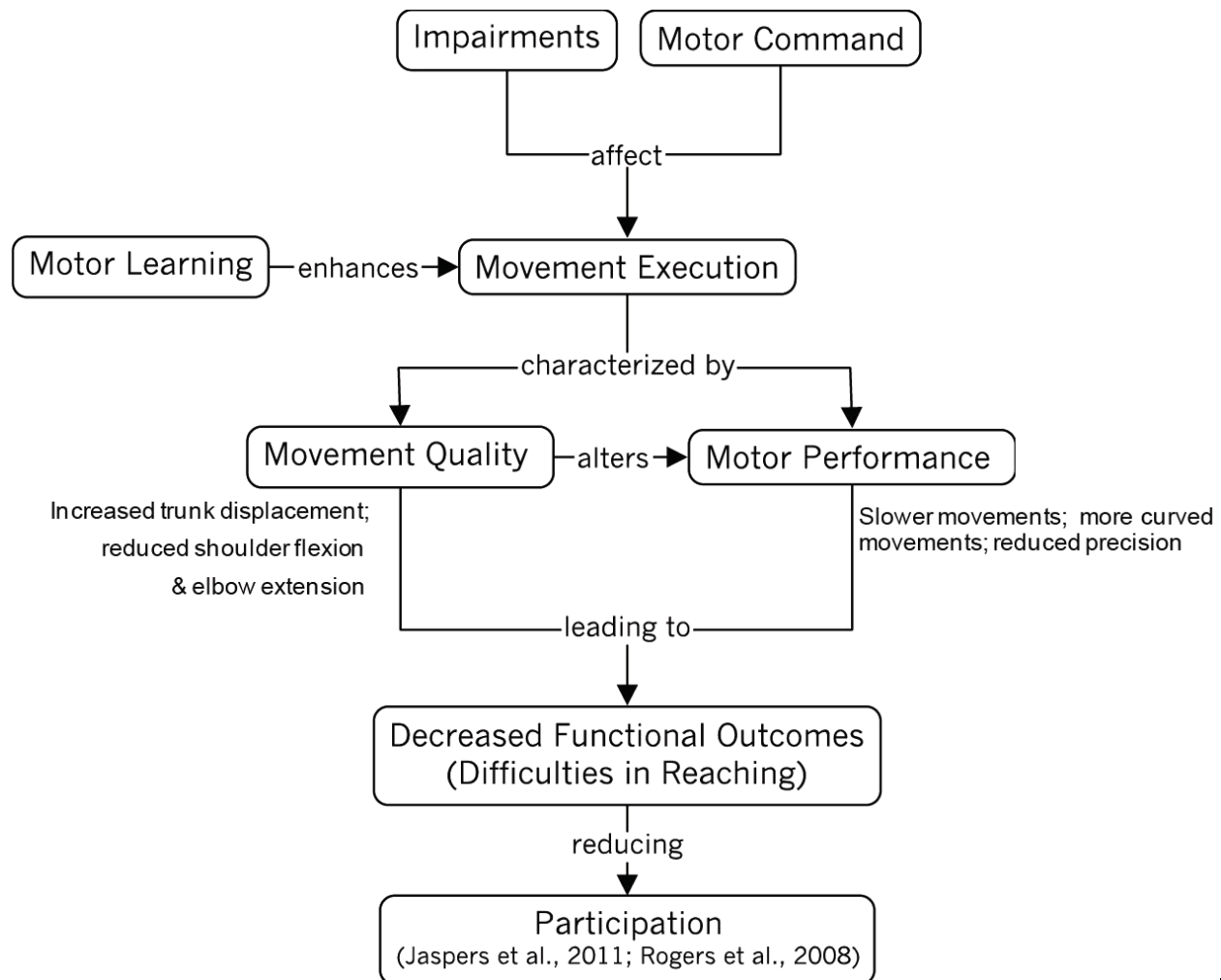


Figure 2-1 Flowchart of the importance of improving movement execution in order to optimize motor learning

2.2 Movement Production

The ability to perform a reaching movement depends on the organization of the cerebral cortex as a whole and the individual's interaction with the environment (Kalaska et al., 1997). The motor cortex is not fully responsible for the plan of action, but rather, it is part of a network of cortical motor areas, where each has a specific role. Neurons in the motor cortex are more complex in comparison to those in the spinal cord as they contribute to several operations such as action planning, sensorimotor integration and execution of motor commands.

2.2.1 The Primary Motor Cortex

The primary motor cortex contains a topographic map of different parts of the human body (i.e., homunculus; Kalaska, 2009; Rizzolatti and Luppino, 2001). Even though the primary motor cortex was one of the first areas to be discovered, there is still uncertainty about its role and how movements are controlled. Studies of primates suggested that the primary motor cortex processes directional information about reaching movements (Georgopoulos et al., 1982; Georgopoulos et al., 2007; Koike et al., 2006). For example, when a monkey reached for targets in different locations, neuronal activity corresponded to the direction of the movement made by the monkey. However, each neuron had stronger activity in a preferred direction reinforcing the idea that the primary motor cortex contributes to the execution of a reaching movement (Georgopoulos et al., 1982; Georgopoulos et al., 2007; Koike et al., 2006).

There is also minor evidence that primary motor cortex neural activity is correlated with the muscle elasticity and force (Caminiti et al., 1990; Gribble and Scott, 2002; Kalaska et al., 1997; Morrow et al., 2007; Sergio and Kalaska, 2003). It is also important to note that other primary motor cortex neurons are not correlated with the electromyographic output of the muscle. However, the concept that movements are guided by the primary motor cortex neural activity has been challenged by scientists who propose that motor commands set parameters that only indirectly result in force output (Feldman and Levin, 2010). According to this theory, movements are not directly specified by the neural activity of the motor cortex, but rather by shifting the thresholds of muscle activity or neuromuscular elements without the need of any computational transformation to generate the required forces (Feldman, 2016)

Moreover, many neurons of the primary motor cortex receive sensory input. This information is hypothesized to be crucial as it characterizes the ongoing movement. Furthermore, the provision of sensory input informs about the errors experienced during any movement. This information is crucial as it facilitates motor learning once the corrections are made after repeated practice. Hence, the primary motor cortex is hypothesized to act as a dynamic neuronal pool converting central signals about the desired action and the sensory information to send a motor output in the

descending pathways like the corticospinal tracts (Kalaska, 2009). However, more evidence is needed to confirm or to refute this hypothesis.

2.2.2 The Parietal and Premotor Cortex

As mentioned earlier, the ability to perform a reaching movement depends on dynamic transformations of sensory information to motor output. Thus, the choice of action for the completion of a desired movement requires sensory information about the environment and the body. For example, reaching an object requires information from the visual cortex about: 1) the object's location in space and 2) the physical properties of that object.

Sensory information coming from the visual cortex is relayed to different parts of the brain, particularly in the dorsal and ventral premotor cortices and supplementary motor area (Cisek and Kalaska, 2010; Haxby et al., 1991). These regions contribute to the integration of visual information, a process defined as “sensorimotor integration” or “visuomotor transformation.” The first step to process visual information occurs in the primary visual cortex. Two major pathways carry information about the objects and the environment (Goodale and Milner, 1992).

The first pathway is the ventral or the “what” pathway, which ends in the temporal lobe, carrying information about object recognition and form representation. Object recognition relies heavily on perceptual constancy. Perceptual constancy is exemplified by the concept of size constancy, i.e., an object placed at different locations in the environment will always be perceived as having the same dimensions even though the sizes may differ (Sperandio and Chouinard, 2015). Hence, object recognition plays an important role in object perception as faulty recognition could result in distortion of the object location.

The second pathway, called the dorsal pathway, also known as the “where and how” pathway, primarily ends in the parietal lobe, carries information on where the object is located in space, crucial information for the guidance of a movement (Yamagata et al., 2009). First described by Gibson (1954), the parietal lobe gathers sensory information about the object, which is described

as object affordances. Examples of object affordances include, but are not limited to the texture and the design of the object..

2.2.3 Classification of Movements

Kinematic analysis of a reaching task can reveal the different strategies that underlie the performance of the desired task. Generally, kinematic analysis provides additional information through spatiotemporal parameters on movement preplanning, execution and smoothness (Chen et al., 2013). However, most studies provide spatiotemporal parameters of the movement task without distinguishing between how the movement is made and what is improved at the performance level (Chen et al., 2013; Wu et al., 2007). Without addressing the quality of the movement, it is not possible to distinguish motor compensations used to accomplish the task. Examples of compensatory strategies used by individuals with stroke and by children with spastic hemiplegic CP include trunk displacement, scapular elevation and shoulder abduction (Levin et al., 2002; Mackey et al., 2006; Roby-Brami et al., 2003; Schneiberg et al., 2010). The overuse of different compensatory strategies in the long-term may lead to injuries such as back problems and occasional joint pain (Ada et al., 1994; Levin and Sveistrup, 2008).

Thus, to identify the compensatory mechanisms and what exactly is learned, movement execution should be described at two levels: 1) movement quality based on body-centered coordinates and 2) motor performance based on room-centered coordinates (Levin et al., 2009). Referring to how the body moves with respect to itself, movement quality is usually measured in the number of degrees moved by a joint. Specifically, movement quality indicates the range of movement of flexion/extension, abduction/adduction or rotation of different joints. Movement quality also refers to spatial and temporal interjoint coordination and muscle activation patterns (Levin et al., 2009). Variables of movement quality contribute to movement of the endpoint (end effector that interact or handles the object), which can be denoted as motor performance variables. Motor performance variables include endpoint velocity, endpoint trajectory smoothness and straightness, usually measured in spatial coordinates (x, y and z; Liebermann et al., 2010). These two levels have been used in previous studies to describe upper limb reaching in children with CP (Mackey et al., 2005; Schneiberg et al., 2010).

2.3 Neuroplasticity

Neuroplasticity is the process by which reorganization in the nervous system occurs either at the level of behaviour, or at the level of anatomy and physiology during an individual's entire lifespan (Pascual-Leone et al., 2005; Sale et al., 2009). Although more prominent in ages up to 25 years, neuroplasticity happens throughout the lifespan due to continuous changes in responses to stimuli (Pascual-Leone et al., 2005). In animal studies, neuroplasticity is linked with functional improvements, such that the repetition of a reaching task in rodents who had a stroke can lead to an increase of dendritic arborisation and of synaptogenesis, and a better functional neuronal network (Friel and Nudo, 1998; Kleim et al., 2002; Monfils and Teskey, 2004). Furthermore, changes in the white matter structure of the corticospinal tract after motor training in healthy adults were observable by diffusion magnetic resonance imaging (Guzzetta et al., 2007). Lastly, in the light of recent evidence, expressions of both growth-promoting and growth-inhibiting proteins were found in the tissues near the lesion, which might help the development of novel axonal connections in both animal and human models (Murphy and Corbett, 2009; Nudo, 2006).

In patients with brain injury, especially children with CP, the process of neuroplasticity is poorly understood (Reid et al., 2011). A better comprehension may facilitate effective rehabilitation in patients with brain injury. Neuroimaging studies showed that motor recovery is associated with a decrease of neuronal activity in the unaffected hemisphere and an increase in activity of the affected cortex after the acute phase in individuals with stroke (Pascual-Leone et al., 2005). However, this finding was challenged in a recent review that reported increased activation in the unaffected hemisphere in order to compensate the loss of activity in the affected hemisphere, possibly contributing to recovery of movement (Di Pino et al., 2014). Other studies have also demonstrated that the increased activity in the intact hemisphere can lead to over-inhibition of the affected hemisphere through interhemispheric inhibition (Murase et al., 2004).

In children with early injury, the immature brain has a large potential to compensate for deficits in motor function. In children with hemiplegic CP, recruitment of ipsilateral tracts originating in the unaffected hemisphere can substitute for the loss of control of the paretic hand from damaged

contralateral pathways of the paretic hand (Carr et al., 1993; Staudt et al., 2002). For example, a comparison of the recruitment of ipsilateral tracts in congenital hemiparesis to age-matched controls using transcranial magnetic stimulation and functional magnetic resonance imaging found that the recruitment of ipsilateral activation from premotor areas was independent of the lesion severity. However, in the group of children with more severe lesions, ipsilateral activation was also found in the primary sensorimotor region of the unaffected hemisphere (Staudt et al., 2002), suggesting that the somatosensory functions can be preserved and that neuroplasticity may occur in children with severe lesion. However, increased recruitment of ipsilateral tracts is also associated with decreased hand function in children with CP when compared to children without recruitment of ipsilateral pathways (Holmstrom et al., 2010). There is still a need for better understanding of the ipsi- and contralateral intra- and interhemispheric interactions that may be affected by pre- or perinatal brain lesions in children. Neuroplasticity after repetitive practice of a motor task results in a reorganization of brain networks and a more efficient functional network in different cortical areas such as the primary motor and the primary somatosensory cortices, supplementary motor area and putamen (Dayan and Cohen, 2011; Kelly and Garavan, 2005; Rotem-Kohavi et al., 2014). For example, intensive bimanual training in children with hemiplegic CP led to an increase in the size of the affected hand motor map and amplitudes of motor evoked potentials (Kuo et al., 2016). Moreover, the greatest changes in hand motor maps in children were associated with greater functional improvements on the Canadian Occupational Performance Measure. In another study, a reorganization of the sensorimotor cortex was observed in children with hemiplegic CP who participated in a 12-day intervention of constraint-induced movement therapy (Juenger et al., 2013). Specifically, functional magnetic resonance imaging and transcranial magnetic stimulation demonstrated an increase in S1 activation, but a decrease of M1 activation in children whose ipsilateral corticospinal projections were not pruned by the age of two (Juenger et al., 2013). In future research, the development of the corticospinal tracts and the interhemispheric interactions should be addressed as it may serve as an explanation of why some children were able to improve, while others were not.

It is important to understand that neuroplasticity could also be maladaptive or as earlier authors suggested, to have a “dark side” (Elbert and Heim, 2001). Maladaptive plasticity refers to brain reorganization that leads to dysfunctional outcomes (Nava and Roder, 2011). For example, the

reinforcement of undesired movement patterns could have a negative effect on motor learning such as the strengthening of undesirable connectivity of different brain regions or the pruning of important connectivity, which could lead to undesired or altered movement. For instance, a highly skilled pianist could develop movement disorders such as focal hand dystonia in response to exhaustive repetitive rehearsal of the skills or dramatic change in technique (Quartarone et al., 2003; Quartarone et al., 2006). Results of a functional magnetic resonance imagery analysis explained this so-called “musician's cramp” as a reorganization of the topographic map in the somatosensory cortex (Tamura et al., 2009). In other words, the representation of the hand is less segregated in both hemispheres of the brain in highly skilled pianists who practiced excessively (Candia et al., 2003). Another example of maladaptive plasticity in the sensory cortex is phantom pain, which occurs in certain cases of amputation (Flor, 2008). The mechanism underlying the phantom pain felt by amputees is poorly understood and may result from a combination of physiological and chemical changes in different regions of the cortex and at peripheral levels (Flor et al., 2006). The pain felt by amputees has been associated with the reorganization of the sensory cortex causing a loss of GABAergic inhibition and/or structural alterations including axonal sprouting. Moreover, the intensity of pain prior to the amputation is predictive of the phantom pain after deafferentation, reinforcing the idea that the memory of pain is an important mechanism to consider (De Ridder et al., 2011; Nikolajsen et al., 1997). Little is known about what leads to maladaptive plasticity, especially when compared to adaptive plasticity.

A recent review about individuals with stroke recognized the possibility of maladaptive plasticity leading to compensatory movement patterns (Jang, 2013; Krakauer, 2006; Lee et al., 2009; Schwerin et al., 2008). Other studies have reported that peripheral factors contribute to compensatory movements (Archambault et al., 1999; Cirstea and Levin, 2000; Roby-Brami et al., 2003). Thus, the concept of maladaptive plasticity is important to take into account when learning a new task in order to avoid the reinforcement of undesirable movement patterns.

2.4 Motor Learning

Improvement on movement quality and motor performance is essential in order to increase a child's participation in activities such as those measured by the Jebsen-Taylor Test and the

Melbourne Assessment (Chen et al., 2013; Chen et al., 2014; Eliasson and Gordon, 2000; Klingels et al., 2012). Over the last 20 years, various therapies (e.g., constraint-induced movement therapy, neuro-developmental therapy, hand-arm bimanual intensive therapy) have been put forth with the objective to either enhance movement behavior or to decrease the functional limitations of children with CP (Gordon et al., 2008; Gordon et al., 2011; Gordon et al., 2007; Novak et al., 2013; Parette and Hourcade, 1984; Snider et al., 2010). However, when an intervention was not as successful as predicted, common explanations included weak methodology, non-optimized settings for the intervention and inappropriate use of principles of motor learning. Since motor learning is fundamental to any motor behavior improvements, the appropriate use of principles of motor learning is critical.

Schmidt and Lee (2011) define “motor learning” as a set of processes associated with practice or experience leading to relatively permanent changes in the capability for movement. Motor learning is based on different principles that should be given special attention in a treatment intervention aimed at improving motor function. In training programs promoting neuroplasticity and learning, characteristics based on motor learning include how frequent the intervention is given, feedback frequency, types of feedback, total practice time and types of practice (Vasudevan et al., 2011). Based on the Schmidt definition, learning consists of four characteristics. The first characteristic is a set of change processes when a task is practiced enabling the learner to become more skilled at it. The second characteristic of learning occurs directly from practice or experience. The third characteristic is that learning cannot be observed directly and has to be measured through changes in behavior i.e., the quality or the performance of the movement. Lastly, the fourth characteristic states that learning leads to relatively permanent change in a movement.

2.4.1 Theories of Motor Learning

Over the past 50 years, several theories have been proposed on how an individual learns a new task, but there is still no consensus as to which theory best describes the phenomenon as each has its advantages and limitations (Muratori et al., 2013). Nonetheless, each theory brings an important perspective of factors to be considered when motor learning is optimized. In the next

section, three different theories are briefly discussed followed by a presentation of principles that could influence motor learning.

2.4.1.1 Schmidt's Theory of Learning

Schmidt's theory of motor learning attempted to challenge and to overcome the limitations of earlier theories such as the closed-loop motor control theory (Schmidt, 1975; Schmidt and Wrisberg, 2008). A main criticism of the closed-loop theory was that if learning constantly relied on the use of continuous intrinsic feedback (sensation), then it could be only applied to simple and slow movements (Sherwood and Lee, 2003). Schmidt proposed the existence of two constructs: the generalized motor program and the schema. The generalized motor program basically contains the invariant features to carry out a movement. In other words, the generalized motor program governs the structure of the movement with information, such as the sequencing of submovements, the relative timing and the relative forces (Shea and Wulf, 2005). Thus, the generalized motor program facilitates the production of new movements based on previously learned movements. When learning a new movement, an individual will either generate a new generalized motor program or modify an existing one. As for the schema, it could be divided into two sections: recall and recognition. The recall schema refers to the selection of parameters chosen by the generalized motor program for a desired movement. The recognition schema associates a generalized motor program and a movement outcome. Hence, if an error is detected, the schema will be modified based on the received feedback, which will ultimately lead to a better performance through the learning process. Schmidt was among the first to introduce the concept of external feedback into motor learning, which is now considered as a fundamental factor.

2.4.1.2 Bernstein's Theory of Learning

According to Bernstein (1967), one of the main difficulties of learning a new task is the problem of redundancy of degrees of freedom. Essentially, motor redundancy refers to the problem of finding a unique solution when there are multiple elements (or degrees of freedom) to consider.

For example, the problem can be seen at multiple levels of the neuromotor systems. This problem was illustrated with a reaching task (Bernstein, 1967). For example, while an object location is described in a 3D space having 3 parameters (x, y z), the number of joint degrees of freedom of the arm exceeds three (e.g. three rotations in the shoulder, two at the elbow, two at the wrist, etc.) allowing an infinite number of possible combinations of these elements to complete the task. The number of ways to make an error increases due to the abundance of degrees of freedom. Recent critics of the problem of redundancy of degrees of freedom suggested that it may be conversely considered as a “bliss of abundance” as the degrees of freedom could be useful for many aspects of motor behavior (Latash, 2000; Latash, 2012a). The principle of abundance states that based on the many available elements, the nervous system will propose different solutions to complete the desired action without altering the accuracy of any action (Gelfand and Latash, 1998; Latash, 2012a). According to Bernstein, learning a motor task requires learning how to coordinate different elements of the body to achieve a task. To describe this learning process, he proposed three different stages: 1) freezing degrees of freedom, 2) releasing and reorganizing degrees of freedom and 3) exploiting the mechanical and inertial properties of the body (Bernstein, 1967).

In the first stage, there is an abundance of degrees of freedom, which increases the complexity and the variability of the action. The movement is organized in a way to freeze most of the degrees of freedom. This freezing limits the number of possibly ways to achieve the task, and simultaneously, the complexity of the action (Vereijken et al., 1992).

In the second stage, improvements of skills are characterized by the gradual releasing of degrees of freedom. This release of degrees of freedom allows a higher level of success as the coordination between different structures improves (Vereijken et al., 1992). However, among others, Newell et al. (2001) argued that the use of more degrees of freedom to improve the performance may be explained by the task constraints and not by a general learning strategy. Task constraints may be defined as any limitation such as the goal of the task, rules that specify the response dynamics of the body and the objects themselves (i.e.: object properties of size, shape, mass; Newell et al., 2001). For example, if an individual wants to reach for a cup of

coffee but there are several objects in the way, the person will have to modify his trajectory, according to the environmental and biomechanical constraints.

The last stage, according to the Bernstein theory of motor learning, occurs when the system learns how to exploit the muscle synergies and the use of passive forces. Taking advantage of the different properties of the limb results in an optimal interaction between external forces and reduction of energy costs (Latash, 2012b).

2.4.1.3 Gentile's Theory of Learning

According to Gentile (1972), motor learning involves two stages: the initial stage, often labeled as cognitive or exploratory, and the later stage. Gentile consistently referred to the first stage as the “getting the idea of the movement,” whereas the later stage is denoted as fixation/diversification.

As described by Gentile (1972), the idea behind the first stage is that the individual develops and organizes a motor pattern in which it interacts with the external environment. Overall, the individual explores a wide variety of movement possibilities through trial and error along with problem-solving. This practice provides the individual with information regarding how to do the movement and how to use the characteristics of the external environment to one’s advantage. At the end of the first stage, the individual has sufficient coordination, allowing his goal to be achieved.

In the second and final stage, the individual learns a new motor pattern, so that he will attempt to maintain consistency and to refine movement characteristics to achieve the goal. Concurrently, the individual reduces the energy spent to complete the desired movement. Gentile’s theory makes a distinction between fixation and diversification as whether the movement skill is considered closed or open, respectively. For the closed skill, the individual aims to refine the movement pattern. With practice, the movement will be stereotyped, which requires less attention to the external environment (Poulton, 1957). Another terminology for the closed skill is “feedback control,” which refers to the idea that feedback is constantly required to correct

movement errors. However, if the movement is considered an open skill, the individual needs to adjust the desired movement according to the conditions of the external environment. In the case of feedforward control (open skill), no provision of feedback is provided, as the sensorimotor loop is not completed because it is considered a fast movement. Therefore, any movement errors due to inappropriate motor planning will not be corrected during the execution. Thus, the provision of extrinsic feedback on the movement results is required for any improvement.

2.4.2 Classification of Motor Learning

Motor learning can be either explicit or implicit (Schmidt and Lee, 2011). Often interpreted as declarative, explicit learning is an active process in which the individual is aware of the task, which involves cognitive processing. On the other hand, implicit learning occurs when the individual is not consciously thinking about the task. It requires less attention and is the most common form of motor learning (Boyd and Winstein, 2001; Cleeremans et al., 1998). One example of procedural learning is learning how to ride a bike, as the individual is not fully aware of the motor actions that are underway.

Motor learning occurs across the lifespan. Although most of the basics of skill acquisition such as reaching and grasping are associated with early life (up to two years old), the notion of neuroplasticity underlies the possibility of motor learning throughout life, regardless of age and life stages.

2.5 Factors Influencing Neuroplasticity and Motor Learning

According to Kleim and Jones (2008), 10 principles should be taken into account during rehabilitation after brain damage. The following table gives a short description of those principles at the cellular level of the brain that are likely to be relevant for rehabilitation after brain damage and their influence on brain neuroplasticity. Other factors influencing neuroplasticity and motor learning, such as motivation, environment in which the task is

practiced and task difficulty (Liljeholm and O'Doherty, 2012; Sale et al., 2009), are discussed after the table.

2.5.1 Ten Principles of Neural Plasticity

Table 2-1 Principles of Neural Plasticity

Principle	Description
1. Use it or Lose it	The lack of use of different brain region will lead to the inactivity of the cells leading to the loss of function in these cells.
2. Use it and Improve it	A training program that involves a specific region of the brain will lead to stronger connections, thus resulting in functional improvement.
3. Specificity	Specific training will lead to specific neuroplasticity
4. Repetition Matters	In order for neuroplasticity to occur, sufficient repetition of the desired task is required to make new connections.
5. Intensity Matters	Intensity of an intervention will lead to neuroplasticity. High-intensity stimulation will lead to more synapses and stronger connections.
6. Time Matters	Specific forms of neuroplasticity will occur at different times during training. Different events that will lead to neuroplasticity precede or depend upon others.
7. Salience Matters	The training experience should be important enough to the participant to induce neuroplasticity.
8. Age Matters	In response to training, neuroplasticity occurs more efficiently in younger brains than aging brains.
9. Transference	Neuroplasticity modeled from one specific training can benefit other behaviours/movements similar to the training.
10. Interference	Within the same circuitry, previously trained neuroplasticity may impede the formation of new remodeling in response to new

stimuli.

Some precision is still needed with respect to factors that influence neuroplasticity and motor learning. For example, it is still unclear what is the minimal effective intensity of practice for children with CP to see a positive effect on motor learning. For the purpose of this thesis, practice intensity is defined as how difficult the task or intervention is. The minimal number of repetitions required to promote neuroplasticity in children with CP also needs more research, as there is little to no evidence based on children with hemiplegic CP. It is however generally agreed upon that more practice is better, as described in the literature about constraint-induced movement therapy and hand-arm bimanual therapy (Gordon, 2011; Gordon et al., 2011; Muratori et al., 2013). In support, one premise of the two aforementioned therapies is based on the principle of motor learning such that an increased number of repetitions (in a single session) and practice (number of intervention sessions over time) will optimize motor learning.

2.5.2 Motivation

Motivation is an important factor to promote neuroplasticity and motor learning. The main reward pathway of the brain processes motivation. This pathway is situated in the striatum, a structure in the basal ganglia. The dorsal striatum itself was found to play a critical role during motor performance (Yin et al., 2005). The ventral striatum is proposed to support both learning and performance (Atallah et al., 2007), though there are studies that challenge this hypothesis (Yin et al., 2005). An association between motivation, motor skill and early learning was demonstrated in several studies (Liljeholm and O'Doherty, 2012). For instance, the activity increase in both ventral and dorsal areas of the striatum is correlated with intensified rewards and an improvement of a physical effort in healthy adults (Pessiglione et al., 2007; Schmidt et al., 2012). Similarly, in a rodent stroke model, the animals were not able to navigate through a maze when the level of motivation was insufficient (Mair et al., 2002). For children with CP, higher motivation has been associated with reduced activity limitations and improvement on various motor outcomes (Majnemer et al., 2010; Tatla et al., 2013). In the Majnemer et al. (2010) study, parents of 74 children completed a questionnaire evaluating the child's motivation and assessed the family functioning through two standard measures. Results indicated that children with

higher motivation were more likely to participate in more activity with the family in comparison to their peers who were less motivated. Results of a systematic review suggested that low motivation might negatively influence a child's functional potential and the effectiveness of an intervention (Majnemer et al., 2010; Tatla et al., 2013).

2.5.3 Task Difficulty

Task difficulty is a principle that has been implicitly integrated in most motor learning theories and in most interventions (Schmidt and Lee, 2011). The appropriate variation of difficulty levels can lead to improved functional abilities in patients (Piron et al., 2009). Guadagnoli and Lee (2004) developed the Challenge Point Framework to describe the impact of task difficulty on motor learning. According to this theory, learning occurs when the task difficulty is sufficient to challenge the learner's skills, and yet remains achievable. In other words, the task difficulty increases proportionally to the performance improvement experienced by the individual. Therefore, the choice of the task difficulty requires precaution as it is directly linked with predicted success. If the task is considered to be too difficult or too easy for the user, it will negatively impact motor learning (Akizuki and Ohashi, 2015). Therefore, the optimal challenge point changes in the same rhythm as the individual's motor skill improves when additional skills to tackle tasks of higher difficulty are acquired leading the individual to learn more during each new challenge. In other words, the progression of skill difficulty is essential to improve motor function (Taub and Wolf, 1997).

One possible way to increase task difficulty is through the manipulation of the different variables of Fitts' Law (Fitts, 1954). According to the Fitts' formulae, if the width of the target is smaller, or the distance between targets is greater, the task will be more difficult, resulting in a longer time to complete the movement (Fitts and Peterson, 1964; Latash, 2012b). On the other hand, if the task requires a fast completion, the task difficulty will be reduced when the distance is shorter and the target is larger. The speed-accuracy trade-off is a compromise between movement precision and velocity. The speed-accuracy trade-off is a good paradigm for motor learning since the difficulty of the task can be manipulated in a standardized way. For example, during a physical activity, it might be important for the movement to be as precise as possible

and as fast as possible to be more successful. An example of progression of difficulty would be to increase the number of obstacles when reaching to a target based on the participant's performance.

2.5.4 Sensation

Among the other factors discussed, sensation also influences motor execution and motor learning. Motor dysfunction experienced by children with hemiplegic CP may result from impaired transmission of the sensory feedback from the impaired hand (Guzzetta et al., 2007). Sensation has also been linked with motor learning in animal studies (Mao et al., 2011) and in healthy adults (Vahdat et al., 2011; Vidoni et al., 2010). For example, when delivering repetitive transcranial magnetic stimulation over the primary somatosensory cortex with the objective to alter sensation, smaller improvements were found in comparison to those without stimulation (Vidoni et al., 2010). In other words, when sensation is altered, the optimization of a motor task or learning a new motor task could be greatly limited. Since children with CP also experience reduced sensation (Clayton et al., 2003; Vidoni et al., 2010; Wingert et al., 2008), the assumption is that motor learning optimization is consequently limited. However, only one study investigated the impact of reduced sensation on motor learning in children with CP (Auld et al., 2012b). The results demonstrated that children who had larger tactile dysfunction were more likely to have poorer performance on tactile perceptual tests and on the Melbourne Assessment and the Jebsen-Taylor Test (Auld et al., 2012a; Auld et al., 2012b). The conclusion of this study highlights the importance of investigating the role of sensation in children with CP on motor learning.

The reduction of sensation, as that experienced by children with CP, potentially contributes to sub-optimal motor learning. On the other hand, the presence of multiple sensory inputs such as the provision of extrinsic feedback in addition to sensation may or may not be detrimental for optimization of motor learning.

2.6 Multisensory Integration

Optimization of motor learning can occur through the integration of multiple sources of information such as intrinsic feedback (e.g., proprioception and tactile threshold) and additional extrinsic feedback, a concept defined as “multisensory integration” (Lickliter, 2011). This integration of different sources of sensory provides a complete representation of the environment and generates the appropriate behavioral response (Simon, 2008). Evidence from neurophysiological research indicates that sensory systems are linked together very early in development and that the brain is organized in a way to facilitate the integration of information across sensory modalities, enhancing a child's perception to respond appropriately (Calvert and Thesen, 2004; Gori et al., 2011). Furthermore, evidence from human brain imaging studies indicates that cortical pathways that were once thought to be sensory specific can actually be modulated by different sensory modalities (Calvert, 2001; Giard and Peronnet, 1999; Macaluso et al., 2000). The development of the various systems does not start at birth, but rather, occurs in a specific sequence during early development, possibly as early as 2 months old (Tobach et al., 1971). This differential timing provides a context in which the development of sensory systems will not compete or interfere with each other. Thus, the ability to use different cues from the multiple senses may optimize motor learning. For example, healthy adults given additional auditory feedback during a visual learning task had greater improvements than those who received only visual feedback (Seitz et al., 2006). Other studies also determined that the combination of two or more sensory modalities may decrease response time and increase target detection accuracy (Gillmeister and Eimer, 2007; Lippert et al., 2007; Press et al., 2004).

Thus, the process generally observed in healthy adults and TD children may be the same for children with CP. Children with CP with reduced sensation may benefit from this process as other sensory systems can take over to synthesize the information from the external world (Dionne-Dostie et al., 2015). Hence, the provision of additional information sources is recommended in order to compensate for the reduced or impaired sensory system observed in children with CP to either promote the optimization of motor learning or to respond optimally to the task (Dionne-Dostie et al., 2015; Seitz et al., 2006; Taylor-Clarke et al., 2004).

2.6.1 Feedback

According to Schmidt et al. (2008), feedback is essential to provide more information to the participants regarding their performance, which increases the possibility of improving skilled movement such as a reach-to-grasp task. Feedback is classified as intrinsic or extrinsic (Schmidt and Wrisberg, 2008). Intrinsic feedback refers to the brain's awareness of somatic information occurring during task performance. Extrinsic feedback is defined as external information provided by a third party regarding how the task was performed or outcomes of the movement (Schmidt and Wrisberg, 2008). Extrinsic feedback is commonly called "augmented feedback" since it enhances the intrinsic feedback. Van Dijk (2005) mentioned the importance of identifying the content, the form and the timing of the augmented feedback in order to optimize motor learning.

Extrinsic feedback can be delivered using different sensory modalities: visual, auditory, haptic or a combination of them (Sigrist et al., 2013). Different types of feedback for motor learning have been used in studies of interventions in different populations such as stroke survivors, children with CP and TD children (Hemayattalab and Rostami, 2010; Molier et al., 2010; Sigrist et al., 2013). However, there is a lack of evidence for which types of feedback should be prioritized for improvements or retention of improvements of upper limb kinematics in children with CP. In a recent review by Molier (2010), the author mentions the difficulties in determining which type of feedback (visual, auditory or haptic) will have the most benefit for the improvement of upper limb performance or quality of movement in stroke survivors. Visual information is proposed to be the most important modality in daily life specifically for perceiving spatial information (Nesbitt, 2003). In most tasks, vision is required in order to perform the movement. Based on this observation, different theories have been identified relating the importance of visual information for learning such as observation or imitation (Sigrist et al., 2013). As for auditory information, some athletes were found to require auditory information in order to better perform in their respective sports since they already mastered visually perceived information (Hermann et al., 2006). Auditory feedback is hypothesized to reallocate cognitive processing and may help avoid overloading visual perception processes (Eldridge, 2006). Haptic feedback is more prominent for sensory integration especially in infants aged under 5 months, because visual processing is

under-developed (Ayres and Robbins, 2005; Sann and Streri, 2007). Despite the important role of haptic feedback during development, there is a lack of evidence and lack of instruction on how to use haptic feedback (Sigrist et al., 2013). Only one study evaluated the effect of the type (visual vs auditory) of feedback on motor learning in stroke. Maulucci (2001) found that adding auditory to visual feedback led to better improvements in the average speed of reaching, the displacement and the path linearity of upper limb movements in stroke participants. However, there is no evidence for whether visual or auditory feedback should be prioritized for the retention of improvements. As there is limited evidence for which type of feedback might be better to maximize motor learning (Molier et al., 2010), a combination of both auditory and visual feedback is recommended.

Augmented feedback can be scheduled as concurrent (during the movement) or terminal (after the movement). Concurrent feedback is delivered throughout the movement and refers to knowledge of performance (KP), which informs the participant about their quality of movement such as range of motion of different joints (e.g., shoulder flexion, trunk displacement and elbow extension) or their performance of the movement as described previously in this section. Terminal feedback provides knowledge of results (KR), which informs the participant about the outcome of the movement. Some feedback on precision and velocity can only be delivered using KR since it cannot be provided during the movement. The effectiveness of combining KP and KR for improving motor learning during a reach-to-grasp task in people with chronic stroke was previously found (Subramanian et al., 2007). The frequency of feedback can vary from continuous to average to faded. Continuous feedback is delivered after every trial. Average feedback is defined as information given after a fixed number of trials. Faded feedback is defined as a reduction of the feedback frequency over the intervention (Schmidt and Wrisberg, 2008). According to Sidaway et al. (2012), more evidence is necessary to describe which type and which frequency of feedback are most effective in interventions to improve motor function in children with CP. The rationale behind the use of faded feedback is that the individual is required to concentrate more and to rely more on intrinsic mechanisms to improve performance. Too much feedback may interfere with learning and retention phase of tasks in healthy adults and TD children (Young and Schmidt, 1992).

2.6.1.1 Feedback in TD Children

In TD children aged between 8 and 13 years old, Goh et al. (2012) showed that when given 100% or continuous delivery of feedback of timing accuracy during a rapid arm movement task, the participant trajectory was faster in comparison to those who received than faded feedback in terms of improving the movement efficiency based on timing accuracy. Furthermore, Sullivan et al. (2008) showed that faded visual feedback was more suitable in young adults (mean age = 25.6 years, range = 22 to 30) compared to children (mean age=10.7 years, range = 8 to 14) for improving performance accuracy and consistency when learning a discrete, coordinated arm movement using a lightweight vertical lever. Sidaway et al. (2012) showed that in 48 TD children (mean age of 10.7 years), retention of improvements on a task of throwing a beanbag was facilitated using visual KR, given at a frequency of 33% of the trials, about the target zone where the beanbag landed. However, in a more complex task (standing while throwing a beanbag), continuous visual KR led to better retention of throwing precision than faded KR. Wulf et al. (2010) showed that 48 TD children between the ages of 10 and 12 years, performing a soccer throw-in task, retained improvements better when given visual continuous feedback of the movement of the ball compared to children who received feedback every three trials.

To summarize, despite some controversy about which frequency and type of feedback is the most suitable for optimizing motor learning in TD children, continuous KR feedback about precision seems to be more effective in TD children aged between 8 to 12 years old than faded feedback.

2.6.1.2 Feedback in Children with CP

Hemayattalab et al. (2010) showed when KR on where thrown darts landed on a dartboard was given every two trials, retention of the throwing accuracy was enhanced in children with CP. However, when given feedback on every trial about results, participants threw darts that were significantly closer to the center of the target in comparison to children who received less or no feedback during the acquisition phase of practice. In children with CP aged between 8 and 16 years old making discrete arm movements, Burtner et al. (2014) showed no significant difference

in accuracy and in consistency of the performance as measured by the root mean square error between children who received visual continuous compared to faded feedback on precision and consistency of the movement. Although there was no difference between continuous and faded feedback, the continuous feedback group demonstrated less error. However, in comparison to TD children, children with CP showed less precision and consistency.

To summarize, there is a need of more robust research to study the different modalities of extrinsic feedback in children with CP.

2.6.1.3 Feedback in Adults with Stroke

Due to the limited number of studies on motor learning in children with CP, some elements of motor learning are summarized in this section based on studies in individuals with stroke. Overall, studies on the frequency of feedback for motor learning showed that results from adults cannot be interpreted in the same way as for children because of differences in motor skill acquisition, learning and cognitive capabilities during development (Sullivan et al., 2008). The pathophysiology of stroke is not the same as CP, but there are some similarities between those two disorders. Both disorders can result in motor and cognitive deficits.

In stroke participants, there is some evidence on the optimal levels of delivery as well as the effectiveness of feedback on motor learning. For example, Winstein et al. (1999) showed that faded feedback as compared to continuous feedback was better to facilitate the improvement of upper limb precision (root mean square error) and consistency in 40 participants with stroke during a horizontal movement using a lightweight horizontal lever. However, the authors cautioned that these findings could not be generalized to all stroke survivors. In a recent review on extrinsic feedback, a lack of evidence in regards of the frequency that should be prioritized in individuals with stroke was mentioned as there is only one study that compared different frequencies (Molier et al., 2010; Winstein et al., 1999). Most of the studies included in this review did not specify the frequency of the feedback. Once again, the optimal type of augmented feedback provided in individuals with stroke is unclear (Molier et al., 2010). Nonetheless, it is important to note that one study done in individuals with stroke found a desirable effect on

providing both auditory and visual feedback for improving linearity of the movement (Maulucci and Eckhouse, 2001). According to Subramanian et al. (2010), there is a need to understand the relationship between motor learning, the type of feedback (KR and KP) and the delivery schedule of feedback. Therefore, a better understanding about which delivery schedules and types of feedback should be used for improvement and change retention in upper limb kinematics is needed for children with CP and TD children.

2.7 Virtual Reality

Manipulation and standardization of the frequencies of extrinsic feedback are possible through the use of VR systems. VR can be defined as “an approach to user-computer interface that involves real-time simulation of an environment, scenario or activity that allows for user interaction via multiple sensory channels” (Adamovich et al., 2009). Over the last decade, the use of VR to deliver treatment interventions has arisen in different populations such as stroke (Subramanian et al., 2013), CP (Levac et al., 2017; Schneiberg et al., 2010; Snider and Majnemer, 2010), and TD children (Lanningham-Foster et al., 2009). VR may serve as an adjunct to conventional therapy in the aim to increase treatment intensity and to modify principles of neuroplasticity in order to optimize motor learning (Galvin and Levac, 2011). For instance, the use of VR systems in an intervention where the objective was to improve movement skills was found to provide high-level of motivation in children with CP (Bryanton et al., 2006; Harris and Reid, 2005; Snider et al., 2010). Another advantage of using a VR system is the possibility to program the task difficulty level, which is often used as a mean of progression in therapy (Crosbie et al., 2007; Kim, 2005). For example, many VR systems offer the opportunity to change the object size and the allotted time to complete a specific task. In addition, it is possible to specify the required distance to complete the desired movement. Lastly, in VR systems, extrinsic feedback can be easily manipulated and standardized to different frequencies (i.e., continuous, faded, etc.) and types (i.e., auditory or visual) (Riva et al., 2006).

2.7.1 Types of Virtual Environments

Virtual environment (VE) displays are an illusion generated by a VR system, in which the user can interact with its environment without the user's physical presence. VEs can range from being total-immersive to partial-immersion and lastly, to non-immersive (Wilson et al., 1997). One of the variances between the different levels of immersion is the level of presence. In a total-immersive VE, the user is completely isolated from the outside world. Immersive VE, defined as the extent to which technology can deliver an illusion of reality (Slater, 2009), can enhance motor learning by adapting specific characteristics of learning such as the provision of extrinsic feedback and task difficulty. Total immersion VE includes all the senses (i.e. vision, touch, etc). Immersive VEs exist in both 2D and 3D systems and can improve the sensation of presence (Schubert et al., 2001). On the other end of the spectrum, non-immersive VEs are usually in 2D and projected on a display located in front of the user. This projection on a display or through a head-mounted display requires the user to interact with the VE through an avatar controlled by the movements of the user. Widely known examples of a non-immersive VEs include the Kinect™, the Wii™, and rehabilitation-specific applications such as the Handtutor (Meditouch, Israel) and the Jintronix Rehabilitation System (Jintronix, Montreal). Some researchers consider these examples as active video games or low-cost game-based VR systems (Levac et al., 2017; Robert et al., 2013). Even though 2D VEs are generally non-immersive, the illusion can be created in which a 3D impression is provided, referred to as a "2D VE with a 3D rendering." Regardless of the rendition type, both immersive and non-immersive VEs require tracking of the user's movement to complete the intended action.

2.7.2 Virtual Reality and Tracking

The quality of the motion-tracking device used with a VR system is an important factor to consider as it affects the user's perception of his own movement and the movement-related accuracy of the avatar. Generally, the quality of the tracking may be different from an expensive VR system (e.g., CAREN), which uses high-precision systems to record movements (e.g., Optotrak, Vicon and Polhemus), to a more accessible one in game-based low-cost VR systems (e.g. Kinect™ camera for the Microsoft games). Before implementing a VR system in rehabilitation, the limitations of using either a high-quality or low-quality motion-tracking device must be weighed as the choice may alter the desired movement of the participant (Levac and

Galvin, 2013; Tao et al., 2013). The limitations and the impact on movement production are further discussed in Section 2.7.4.

2.7.3 Perception in Virtual Environment

As discussed in Section 2.2, movement production is guided by perception of the object location and the user's intended action (Turvey et al., 1977). Hence, movement quality and movement performance are altered because of the different attributes of the viewing environment, particularly in the game-based low-cost VR systems (Kenyon and Ellis, 2014). Indeed, in VEs, perception of the object location may differ from its location in the physical environment due to altered or limited visual cues. Among the cues that affect the user's perception, the resolution of the display medium can have a negative impact on perception, which may alter the movement of the arm (Kenyon and Ellis, 2014). Consequently, to minimize the impact of the display medium, it is important to ensure that the display resolution matches the standard human visual acuity (Febretti et al., 2013). In addition to the display resolution, the viewer's perspective (e.g., the angle at which the user interacts with the display medium) is also a factor that influences the perception of the user. Another factor that influences the user's perception is the provision of proper visual cues (e.g., object size constancy, shadows, drop-lines, etc.; Mon-Williams et al., (2008)). VEs lacking visual cues could lead to altered perception due to misinterpretation of depth and object location (Kenyon and Ellis, 2014). Improper visual cues can also lead to a misinterpretation of the hand avatar or objects, resulting in altered movements (Kenyon and Afenya, 1995).

2.7.4 Movement Patterns in Virtual Environments

As described in previous Sections 2.7.2 and 2.7.3, motion-tracking and the user's perception in the VE could greatly influence movement performance and quality (Liebermann et al., 2012; Tao et al., 2013; Ustinova et al., 2010). Thus, a better understanding on how VEs can affect movement production is needed as they may impact motor learning.

Until now, comparison of movements made in 2D VE and in 3D VE to those made in a similar physical environments focused solely on healthy adults and on individuals with stroke (Knaut et al., 2009; Liebermann et al., 2012; Magdalon et al., 2011; Viau et al., 2004). Results from these adult studies were consistent. VR that uses high-quality motion-tracking resulted in reaching movements that were slower and more curved in comparison to movements made in the physical environment. Differences in the quality of the movement were also observed. In comparison to movements made in the physical environment, movements made in a VE were found to implicate greater ranges of motion of shoulder flexion and of elbow extension and a reduced use of trunk flexion.

However, these results from studies of adults cannot be extended to TD children and children with CP as the object location and the VE may be perceived differently in comparison to healthy adults due to differences in previous experience (Newell and Verhoeven, 2017), memory and developmental stage (Green and Wilson, 2014).

2.7.5 Virtual Reality and Feedback Provision

Despite the observed limitation of differences in perception and movement production between the physical environment and the VE, the use of VR systems have been proposed to enhance motor learning. Advantages of VR systems include the observation of high motivation, the possibility to manipulate different motor learning principles, and most importantly, to standardize the delivery of extrinsic feedback.

Some studies have used VR in children with CP to improve overall fitness and upper limb function (Robert et al., 2013; Schneiberg et al., 2010). For example, children with spastic diplegic CP expended a high level of energy using an active video game console (Wii™; Robert et al., 2013). Another study also found that children with hemiplegic CP were able to improve upper limb kinematics during a reach-to-grasp task following an intensive intervention using a VR system (Schneiberg et al., 2010). However, no study investigated the effect of delivery of different modalities of extrinsic feedback (Snider and Majnemer, 2010). Nonetheless, these

studies demonstrated the feasibility of carrying out an intervention with encouraging results using different VR systems and the possibility to provide extrinsic feedback on KP and KR.

2.8 Rationale

Children with CP have various impairments leading to deficits in functional activities of the upper limbs and decreased participation, particularly because these limitations increase the difficulty of performing reaching tasks. Reaching tasks can be improved through interventions that promote neural plasticity of the brain induced through motor learning processes.

In order for improvement to occur, the intervention must optimize the use of different principles of motor learning. For example, an intervention needs to be challenging enough while providing a high level of motivation, particularly in children. More importantly, provision of extrinsic feedback on KP or KR is required to optimize motor learning. This provision of extrinsic feedback may compensate the observed reduced sensation in children with CP as it provides additional information.

However, there is a lack of evidence indicating which modalities of feedback (*frequency*: continuous, faded, summary; *type*: visual, auditory, haptic; *parameter*: KR, KP) should be emphasized in order to optimize motor learning in children with CP. Limited research on the effect of the different modalities of extrinsic feedback may be attributed to the lack of an intervention that offers the possibility to manipulate and to standardize the delivery of extrinsic feedback. The use of VR systems to deliver an intervention is a solution to study different modalities of extrinsic feedback as it provides numerous advantages. However, the understanding on the possible limitations on the use of VR systems on both motor learning and movement production is needed. Therefore, it is important to develop and to test the feasibility of a research paradigm using a VR system, in which it is possible to manipulate and to standardize the delivery of extrinsic feedback.

CHAPTER 3 - MOTOR LEARNING IN CHILDREN WITH HEMIPLEGIC CEREBRAL PALSY AND THE ROLE OF SENSATION IN SHORT-TERM MOTOR TRAINING OF GOAL-DIRECTED REACHING

3.1 Preface

The main objective of this thesis is to optimize upper limb motor learning through manipulation of extrinsic feedback in children with spastic hemiplegic CP.

As discussed in Section 2.5.4, sensation plays an important role for motor learning. However, the question remains on how reduced sensation observed in children with CP could limit motor learning in comparison to their healthy peers. The two objectives of the present study addressed the question above. The first and second specific objectives of this thesis were addressed in this manuscript. The first objective was to determine if upper limb kinematics in children with CP during a standardized reach-to-grasp task could be learned, retained and transferred to a similar task. We concluded that about 2/3 of children with hemiplegic CP were able to improve and retain upper limb kinematics following an intervention. The second objective was to characterize the role of sensation on motor learning in children with hemiplegic CP. Results indicated that children with better sensation, specifically tactile thresholds and proprioception were associated with the retention of improvements in endpoint velocity. These results are crucial to understand as reduced sensation could lead to sub-optimal motor learning in children with CP. This study identified the need to investigate whether or not the provision of different modalities of extrinsic feedback could be used to enhance motor learning in children with hemiplegic CP. Such investigation would help to clarify the importance of the use of extrinsic feedback to optimize motor learning and to identify the knowledge and gaps of the literature in TD children and in children with CP.

Motor learning in children with hemiplegic cerebral palsy and the role of sensation in short-term motor training of goal-directed reaching.

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Keywords: Sensation, Cerebral Palsy, Virtual Reality, Children

The final version of this paper has been published in *Developmental Medicine & Child Neurology*, Vol 55 / Issue 12, pp. 1121-1128, December 2013.

3.2 Abstract

Aim: Our aim was to determine if improved upper limb kinematics in children with cerebral palsy (CP) during a reach-to-grasp task could be retained and transferred to a similar task. We also characterized the relationship between sensation and motor learning.

Method: We used a prospective, single-participant research design with 16 children (seven males, nine females; mean/median age 8.6/9y; age range 6–11y) with spastic hemiparesis (Manual Ability Classification System levels II–IV). Children were randomly allocated to one of two groups: (1) task-oriented training with or (2) without trunk restraint. The intervention consisted of three 1-hour sessions per week for 5 weeks (total 15h). Evaluations consisted of sensory modalities (tactile threshold, touch, proprioception, stereognosis) and upper limb kinematics during reach-to-grasp of an object located near and far from the body (five assessments: three pre-intervention, immediately post-intervention and 3mo postintervention).

Results: Motor improvements could be retained 3 months after the intervention and transferred to a similar task in children with CP. Proprioception and tactile thresholds were associated with retention of improvements in endpoint velocity ($R^2=0.34$, $F_{2,13}=4.832$, $p=0.027$).

Interpretation: Practice of activities aimed at improving upper limb kinematics led to better learning and retention of movement patterns in children with CP. Our results underline the importance of sensation for motor learning in children with CP.

3.3 Introduction

Motor skill acquisition continues throughout the lifespan. In typically developing children and adolescents, improvements in upper limb motor skills during the first two decades of life are attributable to maturation in both sensorimotor and cognitive systems (Jovanovic and Schwarzer, 2011). We define upper limb motor skill improvements as occurring in both motor performance (endpoint velocity, trajectory straightness, and/or precision) and in movement quality (active joint ranges, and temporal and spatial interjoint coordination; (Subramanian et al., 2010).

The ability to improve and acquire new motor skills depends on motor learning, defined as internal neural and cognitive processes concerning practice or experience leading to a relatively permanent change in performance (Schmidt and Lee, 2011). The demonstration of motor learning implies that changes in motor behavior are retained after the period of skill acquisition. Another feature of learning is that newly acquired skills transfer to other similar tasks. Despite differences in some aspects of learning (e.g. learning rate, feedback frequency; Vasudevan et al., 2011) motor learning principles are similar for most tasks in adults and children and include elements such as intensive and meaningful practice involving active learner participation (Kleim and Jones, 2008). Practice-induced improvements in upper limb movements have been described in typically developing children. For example, children (mean age 10.7y) who practiced horizontal elbow movements using a lightweight lever improved performance accuracy over four 50-trial sessions (Sullivan et al., 2008). Children who received a higher feedback frequency during practice of an upper limb discrete and coordinated movement using a lightweight vertical lever had fewer errors in retention than those who received less frequent feedback (Sullivan et al., 2008).

Children with cerebral palsy (CP) whose upper limb movements are limited by changes in range of motion (ROM), impaired motor control, tone, and sensation, may not have opportunities to develop typical reaching and grasping patterns (Levin and Sveistrup, 2008), resulting in feeding and self-care problems (Gisel, 2008). In the few studies evaluating upper limb motor skill acquisition in children with CP, findings on learning ability are mixed. Children aged 7 to 14 years had more variable directional errors and more difficulty adapting planar arm movements to

changes in an external viscous force-field compared to typically developing children (Masia et al., 2011). Conversely, similarly aged children with CP (6–12y) were able to incorporate verbal and non-verbal feedback to improve motor performance and retain and transfer some gross motor functions described in the GMFM-66 (Bar-Haim et al., 2010).

Sensory feedback is important for learning motor skills in adults (Vidoni et al., 2010) and it is increasingly recognized that children with CP have sensory deficits (Wingert et al., 2008) that can affect movement production (Auld et al., 2012b; Smorenburg et al., 2012). For example, there was a moderate-to-strong relationship between tactile threshold of the fingers (Semmes–Weinstein monofilaments) and upper limb motor performance (Melbourne Assessment) in 52 children with hemiplegic CP (Auld et al., 2012b). Furthermore, children with hemiplegic CP had more difficulty than typically developing children replicating different target arm positions with both impaired and less-impaired arms, especially for large movement amplitudes (Smorenburg et al., 2012). A decrease of proprioception in children with CP could have accounted for the difficulty of reproducing different target arm positions. However, the role of specific sensory deficits in motor learning remains unclear (Vidoni et al., 2010).

While different treatment approaches have been used to improve upper limb function (e.g. constraint-induced movement therapy; (Taub et al., 2011), bimanual intensive therapy; (Gordon et al., 2007)), few studies have identified improvements in movement quality accompanying functional gains and their relationship to sensory deficits. In a recent study of restriction of compensatory trunk movement on improvements in goal-directed reaching in children with CP, Schneiberg et al. (2010) found that trunk restraint promoted greater improvement in upper limb movement quality for reaches to targets closely located, including less compensatory trunk use. However, practice-induced learning and transfer as well as the effect of altered sensation on motor learning, have not been evaluated.

Our goal was to determine whether upper limb kinematic improvements after reaching training in children with CP were retained and transferred to another similar task. Our second goal was to describe the role of altered sensation in motor learning.

3.4 Methods

3.4.1 Participants

Out of 28 children contacted between 2006 and 2010, 16 children aged 6 to 11 years (mean/median age 8.6/9y) with spastic hemiparetic CP from five Quebec pediatric centers participated in the study. Reasons for non-participation included scheduling problems and non-compliance with inclusion criteria. The convenience sample included 11 out of 16 children who participated in the previous study (Schneiberg et al., 2010) as well as five additional children. Parents signed informed consent forms approved by the Centre for Interdisciplinary Research in Rehabilitation (CRIR) ethics committee. Children aged 11 years signed child-assent forms. Children were included if they had impaired motor control in at least one arm, could sit unsupported, understood instructions, had less than 10° elbow or shoulder contracture, had functional vision, and were cooperative. Children were excluded if they had athetosis, chorea or ataxia, traumatic brain injury, pain, medical or surgical procedures in the 6 months leading up to or planned during the study period, or were receiving upper limb occupational or physical therapy.

3.4.2 Study Design

In this prospective single-participant research, children were closely paired by age and Manual Ability Classification System (MACS) level (Table 3-1) and randomly allocated to one of two upper limb training groups by an individual uninvolved with recruitment, evaluation or training. One group practiced upper limb tasks while trunk movements were restricted by two-three-inch wide straps placed across the chest and attached to the chair, the ‘with trunk restraint’ (WTR) group. The straps limited forward trunk displacement and rotation without constraining scapular movement. Age-appropriate trunk movement was allowed, based on norms in typically developing children (Schneiberg et al., 2002). The second group practiced the same tasks without trunk restraint, the ‘no trunk restraint’ (NTR) group. This group also wore straps but these were not attached to the chair. Evaluations were conducted before (three pre-tests), immediately after (post-test) and 3 months after (follow-up) a 5-week intervention consisting of 15 60-minute sessions performed three times per week. Interventions were performed by an occupational or

physical therapist, unaware of clinical and kinematic evaluation results. A single evaluator, blinded to group assignment, completed all outcome assessments.

3.4.3 Intervention

Equipment and training protocols were identical in all treatment centers (Schneiberg et al., 2010). For each session, children sat on a pediatric chair in front of a custom-made upper limb workstation consisting of horizontal and vertical surfaces (Fig 3-1c). Chair and table height were adjusted so that the horizontal surface was at elbow height and feet were fully supported. The vertical surface extended from the trunk to the limit of the child's arm length and was divided into upper and lower sections by a shelf placed at shoulder level.

Each 60-minute training session consisted of 3 minutes of stretching, 20 minutes of activities performed in each of a physical and a virtual (Fig 3-1b) environment (order randomized), 7 minutes' rest, and 10 minutes' practice of a child/family-selected functional activity. Therapists kept work-logs of training sessions. Trunk restraints (Fig 3-1a) were only used during the 40-minute activity period.

In the physical environment, table workspaces were divided into four horizontal and four vertical quadrants in which children practiced task-oriented activities commonly included in therapy. Training was impairment-based, client-centered and consisted of playing with toys or board games with one or both arms. Activities were progressed taking into account child/therapist preferences and clinical goals. Children received verbal feedback about movement quality from the therapist.

For training in the virtual environment, the vertical workspace was replaced by a computer monitor displaying interactive games controlled by arm and hand movements (IREX, GestureTek, Toronto, ON, Canada). Movements throughout the arm workspace were recorded with a webcam and projected into the game scene. Children received feedback about task success through sounds, game scores, and therapist interaction. All training activities in both

environments were done in an arm workspace defined by the child's arm length, measured from medial axilla to wrist crease.

3.4.4 Evaluations

Clinical testing included sensation—hand dorsum tactile thresholds (stereognosis, touch, proprioception) via Semmes–Weinstein filaments (S-W; Lafayette Instruments, Lafayette, IN, USA) – and upper limb passive ROM. S-W values in typically developing children aged 5 to 9 years range from 2.83 to 3.61mm (Bell-Krotoski et al., 1995). For stereognosis, the number of objects placed in the affected hand successfully identified with eyes closed was expressed as a percentage of the total. Light touch (touch) and proprioception were measured on numeric scales (Fugl-Meyer et al., 1975) with maximum scores of 20 and 8 respectively, indicating no impairment. Upper limb ROM of five arm joints was measured on three-point scales where 24 points represented full, painless ROM. Upper limb impairment and function was measured with the valid and reliable Melbourne Assessment test (Randall et al., 2001). The Melbourne Assessment, which was videotaped and independently analyzed, assesses 16 arm and hand movements for ROM, accuracy, fluency, quality, accomplishment, and/or speed on four- or five-point scales for a maximum score of 122 points.

Laboratory testing evaluated arm and trunk movements during two standardized reach-to-grasp tasks. Movements were recorded (Optotrak 3020, 100Hz; Northern Digital, Waterloo, ON, Canada) from 10 infrared-emitting diodes (IREDs) positioned on the arm and trunk, as previously described.¹ Seated children reached to grasp a 2cm³ block placed at two distances proportional to the child's arm length (close target = arm length, far target = 1+2/3 arm length) in the body midline (Fig 3-1d). Seven to 12 trials per target were recorded in randomized blocks. Close target location corresponded to the distance at which most training occurred. The far target tested the ability of the child to reach beyond the arm workspace, a movement that was not practiced during the training in either environment.

3.4.5 Data Analysis

Kinematic outcomes were restricted to those shown to be reliable for describing arm movements in similarly aged children with CP (Schneiberg et al., 2010): two performance (trajectory straightness, endpoint [hand] velocity) and two movement quality (shoulder flexion ROM, elbow extension ROM) measures. Endpoint tangential velocity data were low-pass filtered (cut-off 10Hz) and used to define arm movement start and end times as when the endpoint tangential velocity rose above/fell below 5% peak velocity for at least 50ms. Peak velocity (mm/s) was determined from the endpoint tangential velocity trace. The index of curvature measured trajectory straightness as the ratio of actual endpoint path length to the length of a straight line joining initial and final positions. Elbow angle was calculated using vectors formed by radius and lateral epicondyle IREDs and lateral epicondyle and ipsilateral acromion IREDs where full extension equaled 180°. Shoulder angle was calculated from vectors between IREDs on the lateral epicondyle and ipsilateral shoulder and lateral

3.4.6 Statistical Analysis

A single-participant research design was used because of small sample size and participant heterogeneity. To determine whether changes occurred in each parameter for each child at post-test and follow-up for each target, we used regression with visual trend analysis and computed effect sizes using standard mean differences. A linear regression line was fit through all three pre-test (baseline) data points and a horizontal straight line was extended from the end of the regression through post-test and follow-up data. The number of data points above or below the line was counted for each phase. Between-mean differences of post-test and pre-test values were divided by the pre-test standard deviation to determine effect size. The same was carried out to calculate effect sizes at follow-up, with post-test replaced by follow-up. Effect sizes of 0.20, 0.50, and 0.80 were considered small, moderate and large respectively (Portney and Watkins, 2000). Effect sizes greater than 0.50 identified motor learning and transfer based on previous studies (Schambra et al., 2011; Sullivan et al., 2008). ‘Motor learning’ was defined by retention at follow-up of improvements in performance and/or movement quality variables for the close

target reach-to-grasp task. ‘Transfer’ was defined as improvements in performance and/or movement quality variables at post-test for the far target reach-to-grasp task.

In this mixed-design analysis, χ^2 tests determined whether the number of children showing improvements in each variable was different between WTR and NTR groups. We also described learning and transfer of kinematic improvements in the whole group of children.

For the second goal, relationships between sensory and kinematic variables were assessed using multiple regressions at post-test (close target), follow-up close target (learning), and post-test far target (transfer). Minimal significance levels of $p < 0.05$ were used.

3.5 Results

All children (seven males, nine females; mean/median age 8.6/9y; age range 6–11y) with spastic hemiparesis (Manual Ability Classification System levels II–IV) completed all study phases and complied with the treatment and testing sessions.

3.5.1 Sensory Evaluations

The profile of sensory deficits in the children in each group was mixed (Table 3-1) with deficits ranging from mild to severe.

3.5.2 Performance Outcomes

3.5.2.1 Endpoint Velocity

For the close target, five children in the WTR and four in the NTR group increased endpoint velocity after the intervention. Improvements were maintained at follow-up in two children in WTR and one in the NTR. Among the nine children who improved endpoint velocity, three WTR children and one in the NTR group showed transfer of improvement to the far target at post-test (Table 3-2).

3.5.2.2 Index of Curvature

Five children in the WTR and four in the NTR group improved trajectory straightness for close target after training. Improvements were maintained at follow-up in three children in each group. Among the nine children who improved trajectory straightness for the close target at post intervention five children in WTR and two in NTR showed a transfer of improvement to far target post-intervention (see Table 3-3, online supporting information).

3.5.3 Movement Quality Outcomes

3.5.3.1 Elbow Extension

For close target, six children in WTR and three children in NTR improved elbow extension range after the intervention. Improvements were maintained at follow-up in one child in WTR and two children in NTR and transfer of improvement to far target at post-test occurred in five children in WTR and two in NTR (See Table 3-4, online supporting information).

3.5.3.2 Shoulder Flexion

Four children in WTR and two in NTR improved shoulder flexion range for close target after training. Improvements were maintained for close target at follow-up in three children in WTR and one in NTR. Improvements transferred to far target at post-test for three children in WTR and one in NTR (see Table 3-5, online supporting information).

3.5.4 Overall Learning and Transfer of Improvement Effects

There were no differences between groups (WTR, NTR) in the number of children who learned or transferred improvements for any of the four kinematic variables (v^2 , $p > 0.05$). Therefore, the data were combined for all children. Figure 3-2 illustrates how many children (1) improved

motor performance or movement quality variables for close target at post-test based on effect sizes (large pie charts in each panel); (2) retained improvement for close target at follow-up (left small pie charts); and (3) transferred improvements for far target at post-test (right small pie charts). Approximately two-thirds of the children showed evidence of motor learning and transfer of improvements for each of the four kinematic variables.

Melbourne Assessment scores were correlated with the index of curvature post-test ($r=-0.63$, $p=0.02$) and at follow-up ($r=-0.60$, $p=0.03$) for close target and at post-test ($r=-0.75$, $p=0.004$) for far target.

3.5.5 Correlations Between Motor Learning, Kinematic, and Sensory Variables

For the group, improvements (post-test) in shoulder flexion ROM correlated with improvements in endpoint velocity ($r=0.48$, $p=0.029$) and index of curvature ($r=-0.56$, $p=0.015$) for close target. For motor learning (follow-up), endpoint velocity was correlated with index of curvature ($r=-0.33$, $p=0.021$) for close target. However, correlations between changes in kinematic variables were not evident for transfer of learning (far target post-test).

There was a significant association between proprioception, tactile threshold and retention of the improvement in endpoint velocity for close target ($R^2=0.34$, $F_{2,13}=4.832$, $p=0.027$). However, there was no relationship between index of curvature, shoulder and elbow ROM, and sensation for motor learning.

3.6 Discussion

Learning occurred in some children in both groups but learning was not affected by the type of practice. Improvements were documented in both movement performance and quality. Specifically, movements were faster and straighter and accomplished with greater elbow extension and shoulder flexion. We also found that decreased sensation affected motor learning.

Children in both groups improved kinematics, but there was no difference between groups in terms of retention or transfer. Although a larger number of children in the WTR group showed evidence of learning to make movements faster and with more shoulder flexion, group differences could not be identified because of the small number of participants. It is possible that the use of a trunk restraint is not an important element for learning movement kinematics. However, results are inconclusive and should be interpreted with caution.

3.6.1 Motor Learning

Motor pattern improvements following the 5-week upper limb training program in children with CP were retained for at least 3 months in some children. Although comparable results for motor learning were found in a similar group of children with CP (Schneiberg et al., 2010), this may be the first evidence of transfer of kinematic improvements in children with CP. Motor learning is influenced by many factors, including genetics, somatosensory function (Vidoni et al., 2010), prior experience, and gender (Schmidt and Lee, 2011). Indeed, learning to make faster reaching trajectories was related to better sensation (proprioception and tactile sensation) in our group of children. This supports previous studies on children with CP about the link between these two sensory modalities and motor learning (Auld et al., 2012b; Smorenburg et al., 2012). Indeed, there is ample evidence from animal studies about the dependence of motor and sensory systems for motor production and motor learning (Mao et al., 2011; Vahdat et al., 2011). When a child with CP has decreased sensory information, different systems can be used to guide motor learning. Izawa and Shadmehr (2011) demonstrated that sensory feedback is not solely responsible for motor learning. When reaching during visuomotor perturbations, both sensory and reward prediction errors promoted motor learning, though only the former affected sensory maps. Consequently, using a relevant reward system may improve results during training.

Indeed, differences in learning and retention of other kinematic variables were not accounted for by sensory status alone. These differences could be explained by other factors mentioned above as well as by differences in brain pathologies among the children. Nevertheless, our results support the notion that practice of skilled upper limb movements leads to effective change in kinematics in children with CP (Duff and Gordon, 2003).

3.6.2 Transfer of Improvements

Upper limb performance and movement quality can be improved and changes can transfer to similar tasks following a specific upper limb training intervention. Two-thirds of the participants could transfer improvements to a similar task for each of the four kinematic variables. The transfer of improvements could be explained by the level of motor impairment of the children. Indeed, having less physical impairment may facilitate the ability to improve upper limb performance and movement quality and transfer improvements to a similar task (Bar-Haim et al., 2010). Furthermore, the transfer of improvements may be due to the similarity of the transfer task (Schmidt and Lee, 2011). Improvements in and transfer of learning can also be related to practice intensity or schedule. For example, Bar-Haim et al. (2010) showed that intensive variable practice led to transfer of motor improvements, measured by the Gross Motor Function Measure-66, in children with CP.

3.7 Conclusion

Children with CP can learn to use new motor patterns and retain improvements after practice. Motor skills learned through practice may be transferred to similar movements in children with CP, and those children with better sensory status are likely to be better learners. Future studies should identify how practice elements should be adapted to optimize motor learning in a larger group of children with greater symptom variability. How kinematic improvements may transfer to more functional reaching and grasping tasks should also be addressed.

3.8 Acknowledgements

Thanks are extended to Sheila Schneiberg for some of the data collection. This research was supported by the Canadian Institutes of Health Research. MTR is supported by a Vanier Canada Graduate Scholarship. MFL holds a Tier 1 Canada Research Chair in Motor Recovery and Rehabilitation. The authors thank the children who participated in this study and their parents.

3.9 References

References for this manuscript can be found at the end of the thesis in the Reference section.

Table 3-1 Demographic and clinical parameters of children with CP

SUBJECT	AGE/ SIDE	SEX	MACS	S-W*	STEREO (% recognized)	TOUCH /20	PROPR /8	ROM /24	MELB /122
With Trunk Restraint (WTR) group									
1	9-L	F	4	R=3.6 L=2.8	100	17	8	22	71
2	8-L	F	4	R=2.8 L=4.1	0	19	0	16	56
3	7-L	F	3	R=3.2 L=3.2	83	20	6	24	89
4	9-R	F	3	R=1.7 L=2.8	100	20	5	22	74
5	8-R	M	2	R=3.2 L=3.2	100	20	8	22	N/A
6	11-L	M	3	R=2.8 L=2.4	80	14	8	24	91
7	9-R	M	2	R=2.8 L=2.8	50	10	6	23	N/A
8	9-L	F	2	R=2.8 L=2.4	100	13	8	23	91
No Trunk Restraint (NTR) group									
9	9-R	M	4	R=4.7 L=2.4	20	20	6	24	58
10	10-L	M	3	R=2.8 L=2.4	80	19	6	24	90
11	9-R	F	4	R=3.2 L=3.2	83	16	6	24	77
12	11-R	F	3	R=3.4 L=3.1	57	14	6	23	82
13	8-L	M	2	R=2.8 L=2.4	50	10	7	21	N/A
14	9-R	F	2	R=2.8 L=2.8	80	18	8	23	90
15	6-L	M	2	L=5.5 -	50	13	2	24	N/A
16	6-L	F	2	R=2.8 L=4.3	50	10	7	23	N/A

M, Male; F, Female; L, Left; R, Right; MACS, Manual Ability Classification System; S-W, Semmes-Weinstein; Stereo, Stereognosis;

Propr, Proprioception; ROM, Range of Motion; MELB, Melbourne Assessment.

* For S-W, normal age appropriate range is 2.83 to 3.61.

Table 3-2 Improvement in endpoint velocity indicated by the number of points above the baseline trendline, mean differences and effect size (ES) for reached to the target at arm's length (CT) and the target at arm's length plus 2/3 (FT). Data are shown for two groups of children practicing task-oriented with (WTR) or without (NTR) trunk restraint. Differences are shown for post intervention minus baseline and for follow-up data minus baseline. Moderate to large ESs indicating improvement are shown in bold font and those indicating deterioration are indicated in grey shading.

Velocity	CT Post-Intervention			CT Follow-up			FT Post-Intervention		
Child	Points above base-line trend line	Post minus baseline difference	ES	Points above base-line trend line	Follow-up minus baseline difference	ES	Points above base-line trend line	Post minus baseline difference	ES
WTR									
1	5/11	182.6	1.108	3/10	178.5	1.084	4/11	109.8	0.551
2	5/10	173.6	0.655	1/9	103.3	0.390	4/10	149.1	0.836
3	8/11	203.5	0.858	6/11	86.4	0.364	3/9	281.1	0.784
4	5/10	229.2	0.688	6/9	459.6	1.380	3/11	131.8	0.372
5	10/10	33.1	0.104	11/11	-247.5	-0.774	7/9	62.1	0.223
6	1/10	-281.9	-1.095	5/10	14.5	0.056	8/10	233.6	0.807
7	9/9	340.8	0.835	7/10	8.6	0.021	6/10	-67.9	-0.162
8	6/9	75.2	0.447	0/11	-387.3	-2.303	3/8	6.7	0.016
NTR									
9	4/11	132.0	0.603	0/12	-300.9	-1.374	1/10	-43.0	-0.130
10	6/11	240.2	0.894	1/10	139.3	0.518	4/10	232.4	-0.008
11	7/9	230.0	1.112	3/9	23.2	0.112	7/9	344.2	2.126
12	11/11	-12.7	-0.032	10/10	-166.7	-0.423	7/10	-180.5	-0.453
13	3/7	-149.0	-0.276	5/11	-178.0	-0.330	8/11	365.7	0.752
14	1/10	-341.9	-1.414	7/13	54.8	0.214	0/11	-180.7	-0.619
15	5/10	-66.8	-0.210	0/9	-456.9	-1.438	2/11	-220.8	-0.568
16	6/11	173.6	0.940	0/10	-414.3	-1.303	3/12	-243.2	-0.883

Table 3-3 Improvement in trajectory path straightness (Index of Curvature, IC) indicated by the number of points below the baseline trendline, mean differences and effect sizes (ES) for reaches to the target at arm's length (CT) and the target at arm's length plus 2/3 (FT). Data are shown for two groups of children practicing task-oriented reaching with (WTR) or without (NTR) trunk restraint. Differences are shown for post-intervention minus baseline and for follow-up data minus baseline. Moderate to large ESs indicating improvement are shown in bold font and those indicating deterioration are indicated in grey shading.

IC	CT Post-Intervention			CT Follow-up			FT Post-Intervention		
Child	Points below base-line trend line	Post minus baseline difference	ES	Points below base-line trend line	Follow-up minus baseline difference	ES	Points below base-line trend line	Post minus baseline difference	ES
WTR									
1	11/11	-0.08	-0.682	10/10	-0.02	-0.150	3/11	-0.03	-0.749
2	0/10	-0.07	-0.229	1/9	0.09	0.294	6/10	-0.003	0.020
3	11/11	-0.18	-1.167	9/11	-0.01	-0.044	9/9	-0.10	-0.924
4	3/10	0.17	0.877	4/9	0.21	1.100	3/11	0.10	0.928
5	9/10	-0.16	-0.817	10/11	-0.16	-0.832	3/9	-0.05	-0.644
6	4/10	-0.11	-0.805	5/10	-0.19	-1.354	3/10	-0.11	-0.873
7	7/9	0.01	0.050	4/10	0.18	1.097	5/10	-0.13	-0.666
8	5/9	-0.18	-1.169	2/11	-0.13	-0.811	6/8	-0.24	-0.839
NTR									
9	5/11	0.11	0.541	10/12	-0.15	-0.778	1/10	0.17	1.463
10	10/11	-0.26	-1.255	7/10	-0.20	-0.957	10/10	-0.18	-0.028
11	7/9	-0.20	-1.572	3/9	0.04	0.285	5/9	-0.11	-0.630
12	1/11	-0.08	-0.720	0/10	-0.06	-0.524	1/10	-0.04	-0.701
13	0/7	-0.09	-0.204	9/11	-0.42	-1.004	0/11	0.21	0.824
14	10/10	-0.18	-0.854	12/13	-0.15	-0.697	8/11	-0.11	-0.315
15	0/10	0.36	1.086	0/9	-0.27	-0.824	1/11	0.13	0.357
16	0/11	-0.02	-0.019	0/10	-0.02	-0.029	0/12	-0.09	-0.150

Table 3-4 Improvement in elbow extension indicated by the number of points above the baseline trendline, mean differences and effect sizes (ES) for reaches to the target at arm's length (CT) and the target at arm's length plus 2/3 (FT). Data are shown for two groups of children practicing task-oriented reaching with (WTR) or without (NTR) trunk restraint. Differences are shown for post-intervention minus baseline and for follow-up data minus baseline. Moderate to large ESs indicating improvement are shown in bold font and those indicating deterioration are indicated in grey shading.

Elbow	CT Post-Intervention			CT Follow-up			FT Post-Intervention		
Child	Points above base-line trend line	Post minus baseline difference	ES	Points above base-line trend line	Follow-up minus baseline difference	ES	Points above base-line trend line	Post minus baseline difference	ES
WTR									
1	10/11	4.9	0.806	10/10	9.7	1.592	1/11	-13.3	-0.537
2	1/1	11.1	1.635	0/2	2.6	0.389	2/3	19.8	1.916
3	11/11	9.7	0.975	10/11	1.3	0.133	8/9	11.6	0.829
4	10/10	13.3	1.467	0/9	-17.4	-1.922	11/11	20.9	2.019
5	2/10	-5.0	-0.648	0/11	-6.5	-0.846	1/9	-4.8	-1.014
6	9/10	-0.4	-0.073	10/10	8.1	0.992	10/10	4.5	0.360
7	6/8	6.0	0.523	0/10	-16.8	-1.450	9/9	8.9	0.690
8	2/9	6.2	0.964	0/11	-12.4	-1.954	1/8	5.5	0.553
NTR									
9	-	-	-	-	-	-	-	-	-
10	11/11	26.4	2.115	0/10	3.2	0.254	10/10	28.3	0.225
11	5/9	0.8	0.071	0/9	-21.3	-1.767	0/9	0.1	0.006
12	10/11	19.7	3.75	10/10	25.1	4.793	10/10	14.7	2.014
13	0/7	-2.4	-0.167	0/11	-5.8	-0.412	4/11	16.5	1.039
14	11/11	10.6	0.729	13/13	13.6	0.938	11/11	16.3	3.745
15	0/10	-8.2	-0.933	3/9	3.5	0.397	10/10	19.7	-1.625
16	0/9	-15.9	-3.570	5/10	0.0	-0.002	1/2	-0.9	-0.090

Table 3-5 Improvement in shoulder flexion indicated by the number of points above the baseline trendline, mean differences and effect sizes (ES) for reaches to the target at arm's length (CT) and the target at arm's length plus 2/3 (FT). Data are shown for two groups of subjects practicing task-oriented reaching with (WTR) or without (NTR) trunk restraint. Differences are shown for post-intervention minus baseline and for follow-up data minus baseline. Moderate to large ESs indicating improvement are shown in bold font and those indicating deterioration are indicated in grey shading.

Shoulder	CT Post-Intervention			CT Follow-up			FT Post-Intervention		
Child	Points above base-line trend line	Post minus baseline difference	ES	Points above base-line trend line	Follow-up minus baseline difference	ES	Points above base-line trend line	Post minus baseline difference	ES
WTR									
1	3/11	-0.3	-0.087	9/10	1.9	0.557	0/11	-8.2	-2.309
2	10/10	3.1	0.497	8/9	0.7	0.119	10/10	11.2	1.907
3	11/11	12.3	2.466	11/11	5.2	1.045	9/9	10.4	1.303
4	4/10	4.3	0.856	7/9	5.6	1.121	11/11	11.6	1.887
5	5/10	12.5	1.540	11/11	19.4	2.398	0/9	-3.1	-0.585
6	1/10	-3.6	-1.972	10/10	11.4	6.244	1/10	-6.4	-2.591
7	9/9	14.9	1.973	8/10	1.01	0.134	10/10	14.1	1.273
8	0/9	1.4	0.237	0/11	-12.7	-2.221	0/8	-0.7	-0.088
NTR									
9	7/10	1.3	0.152	11/11	9.9	1.149	10/10	0.6	0.051
10	11/11	10.2	1.955	0/10	-2.7	-0.509	7/10	9.8	0.092
11	0/9	-2.6	-0.189	0/8	1.0	0.072	1/9	8.0	0.584
12	6/11	8.0	1.475	6/11	10.2	1.882	10/10	9.4	2.643
13	1/7	-7.9	-0.778	2/10	-6.88	-0.677	0/11	-4.5	-0.626
14	7/11	-0.5	-0.073	13/13	14.9	2.173	8/11	4.1	0.611
15	0/10	-24.1	-1.494	0/9	-7.9	-0.491	0/10	-38.3	-5.280
16	1/11	-0.1	-0.006	10/10	10.9	1.537	0/2	-14.0	-3.156

Figure 3-1 Upper limb motor training (a, b, c) and experimental (d) set-up for kinematic assessment.

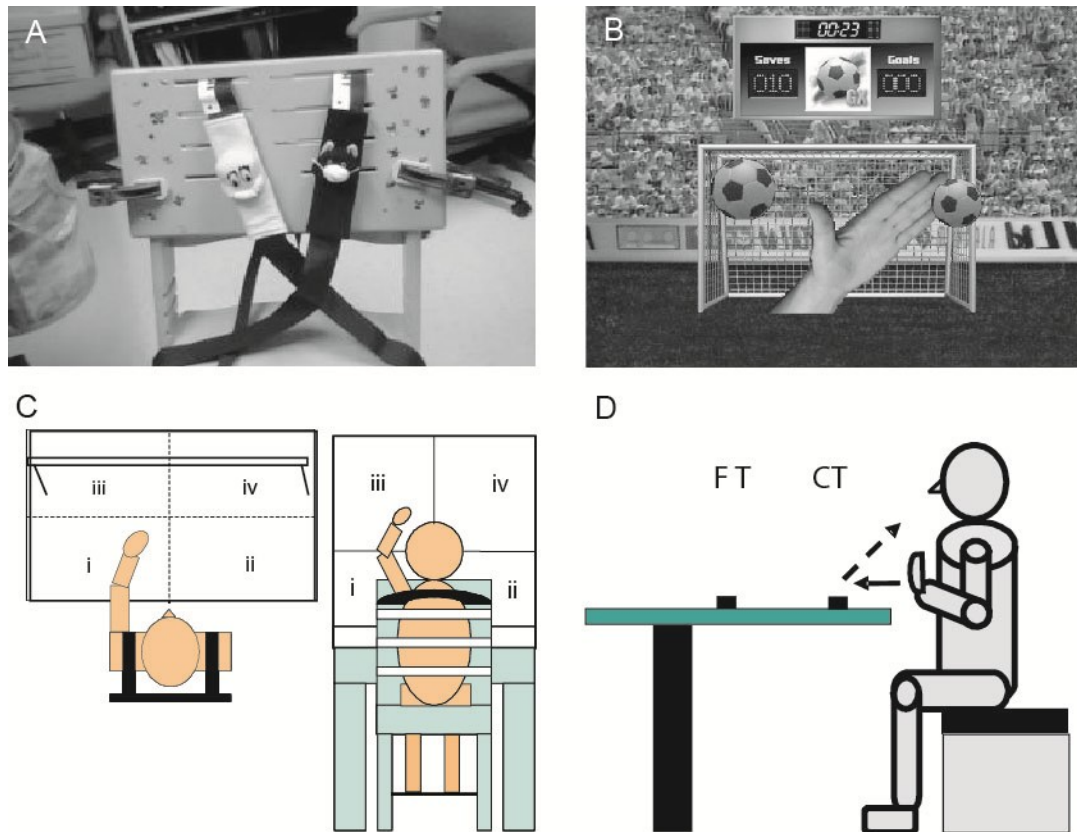
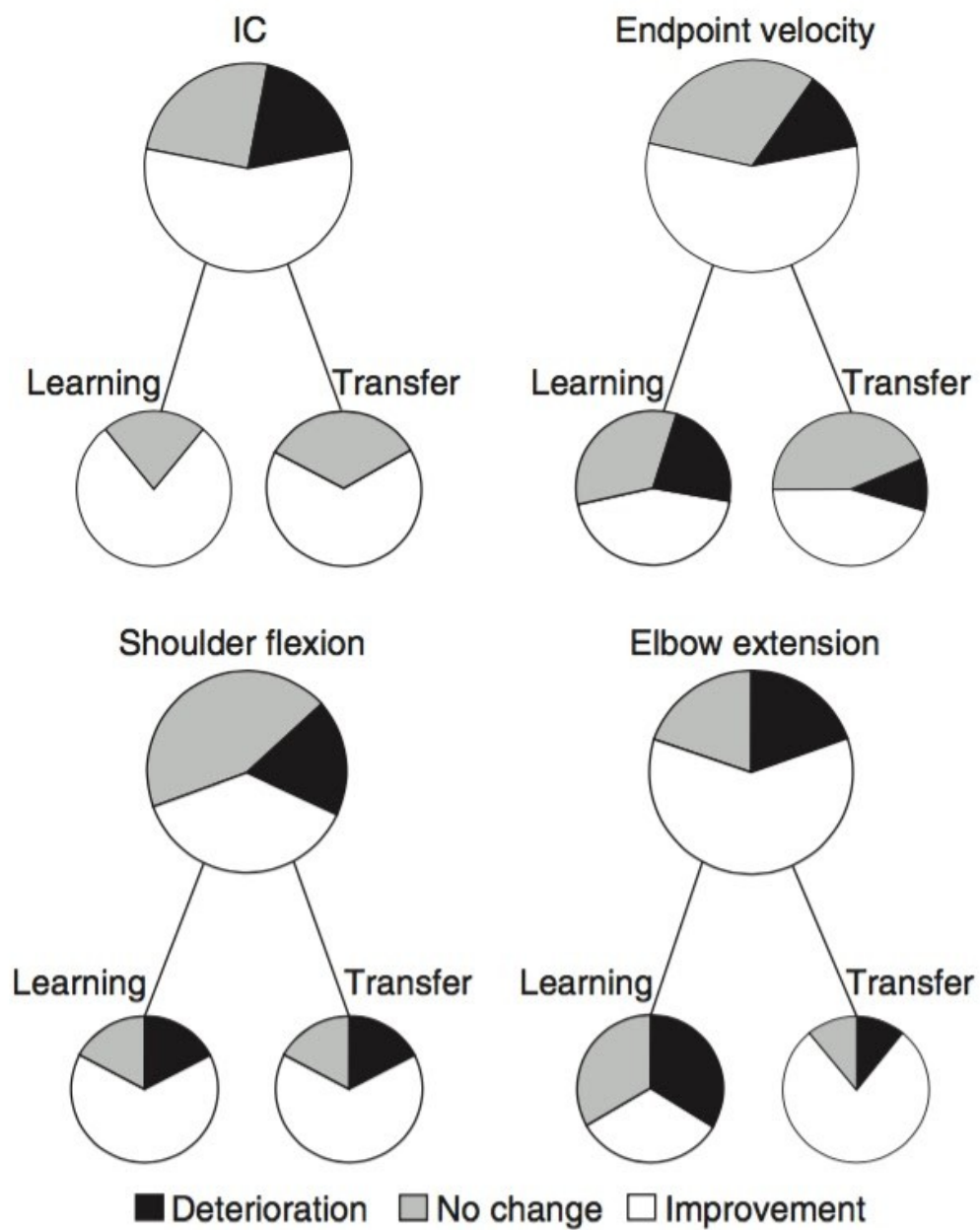


Figure 3-2 Pie charts illustrating the number of children who improved, deteriorated, or did not change on each of the kinematic variables based on effect sizes



CHAPTER 4 EXTRINSIC FEEDBACK AND UPPER LIMB MOTOR SKILL LEARNING IN TYPICALLY-DEVELOPING CHILDREN AND CHILDREN WITH CEREBRAL PALSY: REVIEW

4.1 Preface

As discussed in Section 2.5.4 and in Chapter 3, sensation plays an important role in optimizing motor learning. From the results of the first study presented in this thesis, it is suggested that children with spastic hemiplegic CP who have reduced sensation are limiting the possibility to optimize motor learning.

As children with CP often have impairments that are likely to alter sensation, it is imperative to use a principle of motor learning that could compensate for this limitation. As discussed in Section 2.6, the brain is organized to integrate different information from all sensory systems. Therefore, one possible way to limit the effect of reduce sensation is through the delivery of extrinsic feedback. It is hypothesize that provision of extrinsic feedback will compensate for the reduced sensation and will allow optimization of motor learning. However, the question remains in respect to which modalities of extrinsic feedback should be prioritized, particularly in children with CP.

The objective of this study (Specific Objective 3 of thesis) was to identify knowledge and gaps on the use of extrinsic feedback to improve upper limb motor skills in TD children and in children with CP. This study provides crucial information about the effect of different feedback and helps guide future research and intervention given by clinicians.

Extrinsic feedback and upper limb motor skill learning in typically-developing children and children with cerebral palsy: review

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Keywords: Feedback; children; motor learning; review; upper limb

The final version of this paper has been published in Restorative Neurology Neuroscience, Vol 35 / Issue 2, pp. 171-184, February 2017.

4.2 Abstract

Background: Improving upper limb motor skills occur through motor learning that can be enhanced by providing extrinsic feedback. Different types and frequencies of feedback are discussed but specific guidelines for use of feedback for motor learning in typically-developing (TD) children and children with Cerebral Palsy (CP) are not available.

Objective: Identify the most effective modalities and frequencies of feedback for improving upper limb motor skills in TD children and children with CP.

Methods: Ovid MEDLINE, Cochrane, PEDro and PubMed-NCBI were searched from 1950 to December 2015. English-language articles addressing the role of extrinsic feedback on upper limb motor learning in TD children and children with CP. Nine studies were selected with a total of 243 TD children and 102 children with CP. Study quality was evaluated using the Downs and Black scale and levels of evidence were determined with Sackett's quality ratings.

Results: There was a lack of consistency in the modalities and frequencies of feedback delivery used to improve motor learning in TD children and in children with CP. Moreover, the complexity of the task to be learned influenced the degree of motor learning achieved.

Conclusion: A better understanding of the influence of feedback on motor learning is needed to optimize motor skill acquisition in children with CP.

4.3 Introduction

Motor learning is defined as a set of processes based on principles of neuroplasticity associated with practice or experience leading to relatively permanent changes in a motor task (Kleim and Jones, 2008; Schmidt and Lee, 2011). Motor learning can be enhanced by providing extrinsic feedback to the learner during specific task practice (Kleim and Jones, 2008; Magill, 2011). Feedback is thought to enhance the learner's level of cognitive processing (e.g., attention) and motivation (Laufer et al., 2011; Winstein and Schmidt, 1990). Extrinsic feedback can be delivered through different sensory modalities such as haptic, visual, or auditory information, alone or in combination (Sigrist et al., 2013). Studies suggest that additional auditory or visual feedback contributes to motor learning but inconsistencies between studies suggest further investigation is needed to determine which sensory feedback modalities may be more effective for motor learning (Molier et al., 2010).

Extrinsic feedback can be provided to the learner during (concurrent feedback) and/or at the end (terminal feedback) of a movement in the form of knowledge of performance (KP) or knowledge of results (KR). KP can be provided at two different levels of motor behavior: the motor performance level pertaining to the velocity, precision and straightness of the endpoint, and the movement quality level pertaining to the individual and combined rotations of the joints making up the movement (Levin et al., 2009). In contrast, KR provides information about movement outcomes. KP or KR delivery schedules can be continuous (feedback provided after every trial), average, reduced or summary (feedback after a fixed number of trials), or faded (feedback reduced over time; (Schmidt and Lee, 2011). In adults, frequent feedback enhances movement outcomes during the acquisition phase of learning but improvements may not carry-over into the retention phase (Sigrist et al., 2013). The lack of carry-over of motor gains may result from the dependency of the learner on frequent feedback so that they are less likely to internalize the information for implicit learning to occur (Winstein and Schmidt, 1990). In adults with stroke, no consistent effects of the relative benefits of continuous or faded feedback on motor learning have been reported (Molier et al., 2010).

The complexity of the movement is also an important element to consider when providing extrinsic feedback. Studies have investigated learning of simple movements involving only one degree of freedom that can be mastered in a single practice session (Gottlieb et al., 1988; Wulf and Shea, 2002). However, the coordination of movements of multiple joints is required for the performance of more complex real-world tasks which also require greater use of cognitive processes such as memory, information processing and attention (Wulf and Shea, 2002). A meta-analysis of 40 studies on motor learning of complex tasks in adults showed that continuous extrinsic feedback led to better motor behaviour during the acquisition phase but that faded feedback was more effective for retention (Marschall et al., 2007).

Children with Cerebral Palsy (CP) may use extrinsic feedback differently than adults to enhance motor learning due to impairments in different biological systems. With a prevalence of 1.5 to 2.5 per 1000 births, CP is defined as *“a group of disorders of the development of movement and posture, causing activity limitations that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain.”* (Bax et al., 2005; Paneth et al., 2006). Factors that can affect motor learning in children with CP include sensorimotor impairments, deficits in visual perception (Stiers et al., 2002) and/or deficits in motor planning and cognition (Jones et al., 2007). Besides the existing impairments in children with CP, changes in the developing brain and nervous system should be taken into account when considering guidelines to enhance motor learning. When adults learn a new motor task, neural networks in cortical regions, such as supplementary motor area, posterior parietal cortex and somatosensory cortex are reorganized through remodeling of dendritic spines to incorporate new connections (Hallett, 2005). In contrast, typically-developing (TD) children are constantly learning new motor tasks during development. Children may use information differently for motor learning depending on their stages of sensory, motor and cognitive development compared to adults (Schmidt and Lee, 2011). During infancy, there is a greater reliance on haptic information for motor learning which shifts to auditory and visual information as these systems mature (Moore, 2002; Sigrist et al., 2013). However, how feedback delivered through each of these systems may affect motor learning is unclear.

This review addresses questions about the modality and frequency of additional feedback for upper limb motor skill learning in TD children and in children with CP. In particular, we summarize the evidence for the effect of: a) feedback modality (visual or auditory), b) feedback frequency (faded or continuous) and c) task complexity. The question guiding our review, presented in PICO (population, intervention, comparison and outcome) format was the following: ‘‘ Does provision of extrinsic feedback benefit the improvement and retention of upper limb motor skills compared to no feedback in TD children and children with CP?’’

4.4 Methodology

4.4.1 Search Strategy

An extensive review of the literature published in English was performed by two investigators (MTR, KS). Studies that focused on provision of extrinsic feedback for upper limb motor skill learning in TD children and children with CP were retained. Articles were searched in the following databases: Ovid MEDLINE, Cochrane, PEDro and PubMed-NCBI. Various combinations of MeSH terms and key words were used including feedback, child, children, upper limb, upper extremity, cerebral palsy and motor learning. The search strategy did not impose any restrictions on the year of publication and articles until December 2015 were included. Reference lists of retrieved studies were manually searched to identify other relevant articles.

4.4.2 Selection Criteria

Studies that focused on the effects of different modalities and frequencies of feedback on improving upper limb motor learning in both TD children and children with CP were retained.

Peer-reviewed articles reporting original research were included if they 1) were conducted in TD children or in children with CP under the age of 18 years; 2) assessed improvement in upper limb motor skills through interventions that included the provision of extrinsic feedback regarding

motor performance or movement quality; and 3) compared modalities or frequencies of feedback.

Studies were excluded if 1) children were diagnosed with any other neurological disorders that could interfere with upper limb motor learning, 2) the modality of extrinsic feedback provided was somatosensory (tactile); and 3) the extrinsic feedback provided was on motivation or to improve self-efficacy.

4.4.3 Study Quality Assessment

The retained studies were reviewed for quality by MTR and KS and conflicts were resolved by MFL. Each study was rated with the 27-item Downs and Black Checklist (Downs and Black, 1998), which assesses the methodological quality of both randomised and non-randomised studies based on epidemiological principles. The scale assesses reporting (10 items), internal validity (bias: 7 items, confounding factors: 6 items), power (1 item) and external validity (3 items) of a research study. Each item is rated on an ordinal 2-point scale (0: no/unable to determine and 1: yes) with the exception of two items. The overall quality of the research is scored out of 32. Based on the Downs and Black score, studies were rated as good ($\geq 60\%$), fair (40-59%) or poor ($< 39\%$; Foley et al., 2006). Each study was also rated using the PEDro scale developed by the Centre for Evidence Based Practice in Australia (Moseley et al., 2002). This 11-item scale is based on core criteria generated by expert consensus for assessment of RCT quality. Points are allocated for blinding methods, randomization, data reporting and data analysis. The PEDro classification is the following: 9 to 11, *excellent*; 6 to 8, *good*; 4 to 5, *fair*; and < 4 , *poor* (Foley et al., 2006).

Sackett's quality ratings adapted to include Downs and Black and PEDro ratings determined the levels of evidence for using different modalities and frequencies of feedback for motor learning in children for all studies reviewed (Sackett, 2000). Sackett's levels of evidence are scored on a 7-point ordinal scale, with each level indicating the strength of evidence. A 1a level of evidence was given when the effect was supported by at least one well-designed meta-analysis or two or more good-to-excellent quality randomized control trials (RCTs). If one good-to-excellent

quality RCT was found, a 1b rating level was assigned. A rating of 2a indicated one or more fair-quality RCTs whereas a level of 2b was assigned if there were one or more low-quality RCTs. A rating of 3 was given if the evidence was supported by several pre-post design studies with similar results and a level of evidence of 4 indicated case series studies or poor cohort case-controlled study. Finally, a level 5 denoted the absence of experimental support.

4.5 Results

4.5.1 Data Retrieved

The search strategy retrieved a total of 128 articles, of which 8 met the inclusion criteria. One additional study was retrieved from the reference lists of retained articles. The PRISMA flowchart (Figure 4-1) illustrating the process of selection of articles is appended. These studies included a total of 234 TD children, 102 children with CP as well as 39 young adults.

The quality ratings of the studies based on the Downs and Black checklist are shown in Table 4-1. Seven out of the 9 studies were rated as good and two received a rating of fair.

The Sackett quality ratings to determine the levels of evidences using different modalities and frequencies of feedback are shown in Table 4-2.

The following sections summarize the different modalities and frequencies of extrinsic feedback provided to enhance motor learning in various tasks in TD children and children with CP. Details of the 9 studies are listed in Table 4-3.

4.5.2 Use of Feedback for Motor Learning in Children

4.5.2.1 Feedback Modality

Only one study was found that compared the use of feedback compared to no feedback or another feedback modality for improving accuracy of a throwing task in TD children. TD

children who received continuous extrinsic KR on throwing accuracy improved their scores of precision compared to those who did not receive additional feedback (Abadi et al., 2014). However, this study did not specify the sensory modality used to deliver the feedback, hence no conclusion can be made about whether providing visual or auditory feedback was better for enhancing accuracy during the throwing task.

Two studies were found in children with CP (Sackett's level 3 evidence) showing beneficial effects of additional sensory feedback during the acquisition phase on improving accuracy of throwing darts (Hemayattalab and Rostami, 2010) and a line-tracing task (Talbot and Junkala, 1981). Hemayattalab et al. (2010) provided visual KR at the end of the throwing task whereas Talbot et al. (1981) provided auditory feedback based on errors during the task i.e. when the stylus deviated from the line during tracing. Despite the beneficial effects of additional sensory feedback during the acquisition phase, no effect was observed in the retention phase in both of the studies.

4.5.2.2 Frequency of Feedback in TD children

There is a Sackett's level 2b evidence based on 6 studies that continuous visual and/or auditory feedback is better for improving upper limb motor skills in the acquisition phase but faded feedback is better for retention in TD children.

The difference between additional visual continuous (100%) and faded feedback (62%) on performance accuracy and consistency in TD children practicing a discrete, coordinated arm movement using a lightweight vertical lever was investigated by two groups (Goh et al., 2012; Sullivan et al., 2008). In these two studies, participating children had to replicate the target trajectory using their arm by doing two consecutive elbow extension-flexion movements of specific amplitudes. In each study, groups receiving continuous feedback improved accuracy and reduced variable errors more compared to those receiving faded feedback.

Continuous or reduced feedback had beneficial effects in four other studies in TD children. In two studies, visual and auditory feedback-related improvements were assessed at the motor

performance and movement quality levels during a soccer ball throwing task (Weeks and Kordus, 1998; Wulf et al., 2010). Wulf et al. (2010) showed no significant differences in improvements in whole body movement pattern characteristics (movement quality) during a soccer ball throwing task. At the transfer phase, children who received continuous (100%) feedback performed better in comparison to the reduced feedback group (33%). In contrast, Weeks and Kordus (1998) reported that 33% KP feedback was better for improvement of movement pattern characteristics in the acquisition, retention and transfer phases compared to continuous feedback for the same throwing task. However, neither study found improvements in performance (throwing accuracy), at the acquisition, retention or transfer phases. In another study using a beanbag-throwing task, no significant difference was observed in the acquisition phase regarding the improvement of performance accuracy (Sidaway et al., 2012). However, the retention of improvements of throwing accuracy was facilitated using reduced (33%) visual KR feedback in comparison to continuous (100%) feedback (Sidaway et al., 2012). Similarly, Abadi et al. (2014) showed an improvement of accuracy in the acquisition phase with continuous (100%) feedback during a tennis ball-throwing task in comparison to children who received no or reduced feedback (50%). However, TD children in the 50% feedback group had better throwing accuracy compared to the other groups, in the retention phase.

4.5.2.3 Frequency of Feedback in Children with CP

Based on three studies, there is Sackett's level 3 evidence that either continuous or faded visual feedback delivery can improve upper limb motor learning in children with CP. Hemayattalab et al. (2010) showed that when given KR feedback on the landing location of a thrown dart every trial (100%) during the acquisition phase (5 blocks of 10 trials over 4 sessions), children with CP improved throwing accuracy compared to those who received less (50% KR) or no feedback (0%). However, only the group who received reduced feedback improved accuracy in the retention phase.

Using the same practice schedule, two studies investigated the differences between continuous (100%) and faded (100%, 75%, 50%, 25%) visual feedback on motor performance (precision and consistency) of figure tracing in children with spastic hemiplegic CP (Burtner et al., 2014;

Sullivan et al., 2008). Overall, compared to TD children, children with CP had less precise and less consistent movement profiles but there was no difference between continuous and faded feedback for either group at acquisition or retention phases.

4.5.2.4 Use of Feedback and Task Complexity

There is Sackett's level 3 evidence that either continuous or faded feedback delivery can improve motor learning for complex tasks in TD children based on two studies.

De Oliveira et al. (2009) studied the difference between learning two tasks in TD children (mean age = 11.8 year), where one task was more complex than the other. The latter consisted of throwing a ball with a backward-forward pendular arm movement. The more complex task had the participant perform the same pendular movement followed by an overhead circular movement of the arm. Visual KR delivered at 4 frequencies (25%, 50%, 75%, 100%) was provided on arm performance (throwing accuracy). For each task, the 25% feedback group had more improvement and better retention of the performance based on the number of errors compared to the three other feedback groups. When comparing the tasks, there was no difference in performance between groups. In contrast, Sidaway et al. (2012) reported that continuous visual KR led to better retention of precision gains than faded KR in a more complex beanbag-throwing task (while walking) compared to standing in a similar group of children.

4.6 Discussion

Results from nine studies involving small numbers of participants provide low to moderate evidence suggesting that TD children and children with CP are able to use extrinsic feedback for improving upper limb motor performance and movement quality despite their ongoing development or impairments, respectively. Overall, the studies reported in this review were considered good based on the Downs and Black checklist and the PEDro scale. However, results of the quality of the research evaluation indicate the need for more rigorous and alternative research designs that address different questions such as comparative efficacy.

4.6.1 Feedback Modality: Auditory versus Visual Feedback in TD Children and Children with CP

Only three studies examined the differences between visual and auditory feedback on upper limb motor skill learning in TD children and children with CP. Both modalities of feedback led to better motor learning outcomes compared to no feedback on different scales (Abadi et al., 2014; Hemayattalab and Rostami, 2010; Talbot and Junkala, 1981). Although it is likely that better learning would occur with visual feedback because of the greater spatial information contained in visual compared to auditory signals (Witten and Knudsen, 2005), there is no evidence to suggest that visual feedback is superior or inferior to auditory feedback.

4.6.2 Feedback Delivery: Faded vs Continuous Feedback in TD Children and Children with CP

Evidence indicates that faded feedback about motor performance or movement quality variables may be more effective than continuous feedback for retention of learned upper limb motor skills in adults (Sigrist et al., 2013; Winstein and Schmidt, 1990). Our results suggest that this conclusion may not be directly applicable to children. Based on a total of six studies, there is limited evidence indicating which frequency of feedback may be more effective in children.

Learners who receive continuous feedback about motor performance or quality variables may rely more on this information to improve specific aspects of the movement from one trial to another, without needing to find a set of motor solutions themselves. According to Bernstein (1967), motor learning occurs through the exploration of the relationship between a movement and the physical environment. What the nervous system needs to learn is how best to combine the large number of joint rotations (i.e., kinematic degrees of freedom) to complete the desired movement (Latash, 2012a). The key point is that rather than a single ideal combination of joint rotations, there is a set of movement solutions based on the kinematic abundance of the system (Bernstein, 1967; Latash, 2012a). The idea of learning movement through exploration of the environment was applied to early motor learning in children by Thelen (1994).

Provision of continuous feedback limits the opportunity for learning to occur through exploration, since the learner is provided with specific instruction about how to perform the task on each trial, and may become dependent on it. Provision of faded feedback may result in the system having more opportunity to discover and internalize new movement patterns based on a self-regulatory strategy and retain the new motor skills in the retention stage (Hemayattalab and Rostami, 2010; Meichenbaum, 1986). Despite the reported benefits of faded over continuous feedback for motor learning, in the six studies examining feedback frequency, no conclusion can be drawn about the superiority of one frequency over the other for learning of upper limb motor skills in children. Inconsistencies between studies in outcome measures also makes it difficult to determine which frequency of feedback should be prioritized for upper limb motor learning.

4.6.3 Task Complexity

Continuous visual feedback has been suggested as being more suitable in early learning for a complex movement in children (Marchal-Crespo et al., 2013; Sigrist et al., 2013) since it requires a lower cognitive load when learning involves the integration of a large amount of information (Wulf and Shea, 2002). Indeed, the question of task complexity and cognitive loading takes on greater importance when evaluating motor learning in children with CP who might have associated cognitive impairment. According to Jones et al. (2007), more than two thirds of the children with Cerebral Palsy have cognitive deficits. Given the high prevalence of cognitive impairment in children with CP, more research is needed on the roles of different modalities, frequencies and delivery schedules of feedback with respect to task complexity on motor learning in this population.

4.7 Study Limitations

Overall, the interpretation of the results of these studies is limited by the small number of studies available and the lack of standardization in the interventions and assessments used. None of the studies reported which features of movement were learned based on objectively measured kinematics, which could have provided additional information about the performance and the quality of the movement. For example, a more detailed analysis of the motor patterns would help

distinguish between motor performance and movement quality variables to understand the mechanisms underlying improvements and identify the occurrence of undesirable motor compensations.

4.8 Conclusions

Delivery of appropriately-timed extrinsic feedback has been identified as important for upper limb motor learning in healthy adults and in adults with stroke (Kleim and Jones, 2008; Sigrist et al., 2013; Subramanian et al., 2010). However, it is still unclear what modality type and how much extrinsic feedback should be delivered to maximize learning of functional upper limb motor skills in TD children and in children with developmental disorders, such as CP.

The delivery of extrinsic feedback is thought to be important for improvement of upper limb motor learning in TD children and children with CP. Although all of the studies showed improvement of upper limb motor skills when providing extrinsic feedback, there are contradictory effects of using different modalities and frequencies of feedback. Questions that have arisen from this review indicate that further studies are necessary to identify optimal combinations of feedback modalities, frequencies and delivery schedules as well as motor elements to target in training interventions to improve upper limb skills in TD children and children with CP. However, researchers and clinicians need to be cautious when interpreting results of different studies as they do not necessarily apply to every neurological disorder or every child since CP is a highly heterogeneous condition. Ultimately, it is important to identify how feedback can be optimized for motor learning as well as the role of other principles such as motivation on motor learning (Kleim and Jones, 2008). The complexity of the motor task also should be considered when studying the effect of feedback on motor learning, especially in children with cognitive disorders. Lastly, in order to optimize learning efficiency of children with CP, consideration of the stage of neurological maturation of the child should be considered in the selection of feedback modalities and frequencies. Greater understanding of these parameters and their interactions will help establish better guidelines for designing training paradigms for optimizing motor learning in children.

4.9 References

References for this manuscript can be found at the end of the thesis in the Reference section.

Table 4-1 Study quality determined by 1) Downs and Black Checklist based on percentage scoring $\geq 60\%$, 40-59% or $< 39\%$ (Good, Fair, Poor, respectively), and 2) PEDro scale based on the following criteria: 9 to 11, 6 to 8, 4 to 5, or < 4 (Excellent, Good, Fair, Poor).

Study name	Downs and Black Score /32	PEDro Score /11	Quality of study
Talbot et al., 1981	19	7	Fair
Weeks & Kordus 1998	24	7	Good
Goh et al., 2002	22	8	Good
Sullivan et al., 2008	25	8	Good
Hemayatallab et al., 2010	18	8	Fair
Wulf et al., 2010	20	8	Good
Sidaway et al., 2012	24	8	Good
Abadi et al., 2014	23	8	Good
Burtner et al., 2014	24	8	Good

Table 4-2 Sackett's quality ratings on the different modalities and frequencies of feedback in both typically-developing children and children with cerebral palsy. The number of studies and Sackett rating for each modality/frequency is indicated

Modalities/frequencies	Number of studies	Sackett Rating
Feedback modality in TD children	1	-
Feedback modality in CP children	2	3
Frequency of feedback in TD children	6	2b
Frequency of feedback in CP children	3	3
Feedback and task complexity	2	3
CP: Cerebral Palsy; TD= Typically-Developing.		

Table 4-3 Details of the nine studies retrieved.

Author , year	Populati on	Study design	Intervention	Task compl exity	Feedbac k provided KP/KR	Type of feedba ck visual/ auditor y	Feedback delivery schedule none/faded/ continuous	Outcome measures	Assessment time points	Results
Abadi et al., 2014	45 TD pre- element ary school students Gender not specifie d	Rando mized study – low quality RCT	60 throws (6 blocks of 10 throws) of a tennis ball from shoulder height to a 10 cm diameter target located 3m sagittally; Target consisted of	Simple	KR: not clearly defined	Not availab le	Group 1 – Continuous (100%) feedback Group 2 – Reduced (50%) feedback Group 3 – no feedback	Total score at pre-test, acquisition and retention	Retention test 48 hrs after acquisition	Group 1: better scores during acquisition phase(effect size $r = 0.55^{**}$). At retention, Group 2 had better scores at retention(effect size $r = 0.72^{**}$).

			10 concentric circles indicating accuracy scores							
Hemayat et al., 2010	24 children with spastic hemiplegic CP; aged 7-15 yrs Gender not specified	Pre-post design	30 trials (6 blocks of 5 trials) of dart throwing practiced for 8 sessions	Simple	KR: based on distance from target zone	Visual	Group 1 – Continuous (100%) feedback Group 2 – Reduced (50%) feedback Group 3 – no feedback	Mean accuracy scores	Pre-test, post-test immediately after acquisition and 72 hrs follow-up	Group 1: better scores at post-test compared to Groups 2 & 3 (effect size = 0.87**). Group 3: worst performance. Group 2: better scores at follow-up than Groups 1 and 3 (effect size = 0.78**).
Talbot	59	Rando	40 tracing	Simple	KR:prec	Audito	Group 1 –	Total score of	Pre-test, post	Group 1: higher

et al., 1981	children with CP (spastic, mixed ataxic and athetoid) ; 32 boys; aged 7- 21 yrs	mized study - low- quality RCT	patterns using an infrared stylus; 10 minute training sessions x 2 times/day for 40 sessions		ision of line tracing	ry	error-based feedback when stylus deviated from tracing line Group 2 – no feedback Group 3 – no tracing, no feedback	Southern California Motor Accuracy Test (SCMAT); measuring accuracy and speed	test, 3 mo follow-up	SCMAT scores at post-test compared to Groups 2 and 3. Groups maintained previous performance levels but no differences at follow-up. Effect size is not available.
Goh et al., 2012	19 young adults; mean age 25.6 yrs	Rando mized study - low- quality RCT	Motor task involving moving a lightweight lever 200 times (50 trials/session	Simple	KR: 1. Numeric al total score 2. RMSE 3.	Visual	Group 1 & 3–Adults & Children: Continuous (100%). Group 2 & 4 – Adults	Global movement pattern error (RMSE) –and parameter error (scaling error) Timing factor –	Delayed retention test 24 hrs after acquisition phase	Group 3: better timing compared to Group 4 and 1(effect size $d=$ 1.77*). Spatial

	19 TD children; mean age 10.7 yrs Gender not specified		) to learn specific discrete movements with a specific remembered spatiotemporal goal		Graphic representation of response superimposed on target pattern		& Children: Reduced (62%); feedback gradually faded (100%, 75%, 50%, 25%).	ratio of movement time to target movement time Amplitude factor – best fit trajectory compared to target trajectory		parameters were not affected in any group.
Sullivan et al., 2008	20 young adults (12 males); mean age 25.6 yrs	Randomized study - low-quality RCT	200 (50 trials/session) discrete coordinated reversible flexion/extension elbow movement using a	Simple	KR: RMSE KR: Graphic representation of subject response	Visual	Group 1 & 3–Adults & Children: Continuous (100%) Group 2 & 4 – Adults & Children: Reduced	Performance accuracy (RMSE), consistency (Variable Error)	24 hrs post for retention (no feedback) and reacquisition (feedback received as per group assignment)	Adults: fewer errors compared to children. Group 2 had better performance than Group 1 (effect size = 0.51*).

	20 TD children (12 boys); mean age 10.7 yrs		lightweight lever		superimposed on target pattern		(62%); feedback gradually faded (100%, 75%, 50%, 25%)			Children: Group 3 better accuracy and lower errors than Group 4 Group 4: retention phase better performance than Group 3 (effect size = 0.63*).
Weeks & Kordus 1998	34 TD boys from a basketball summer camp; mean	Randomized study - low-quality RCT	30 trials of throw-ins of a regulation soccer ball to a 2.5 m ² target (cone) placed at 75%	Simple	KR: score around primary target zones KP – movement	Visual and auditory for KR Auditory for	Group 1 – Continuous (100%) feedback Group 2 – Reduced (33%)	Throwing accuracy and movement patterns (form score) specified by 8 skill patterns obtained from	Immediately after acquisition, 24 hrs and 72 hrs post as a retention and transfer test to a target at	No significant differences between groups on accuracy scores at any time point. Group 2: greater form score at all

	age 12.3 yrs		maximal throwing distance; Scoring based on zones around primary target.		nt patterns	KP		videotape analysis	50% of the pre-test distance	three time points compared to Group 1 (effect size = 1.39; 1.79 and 1.19*).
Wulf et al., 2010	48 TD children (18 boys,); aged 10 to 12 yrs	Quasi-random study	30 trials of throw-ins of a regulation soccer ball to a 2.5 m ² target (cone) placed at 75% maximal throwing distance;	Simple	KR – score around primary target zones. KP – movement patterns based on	Visual for KR Auditory for KP	Group 1 & 2 – internal-focused continuous (100%) or reduced (33%) feedback on movement pattern Group 3 &	Maximal throwing distance, accuracy and movement patterns (form score) obtained from videotape analysis	Retention and transfer tests 24 hrs after acquisition phase	Groups 3 & 4 performed better than Groups 1 & 2. Group 3 had better movement form across all time points compared to Group 4. No differences

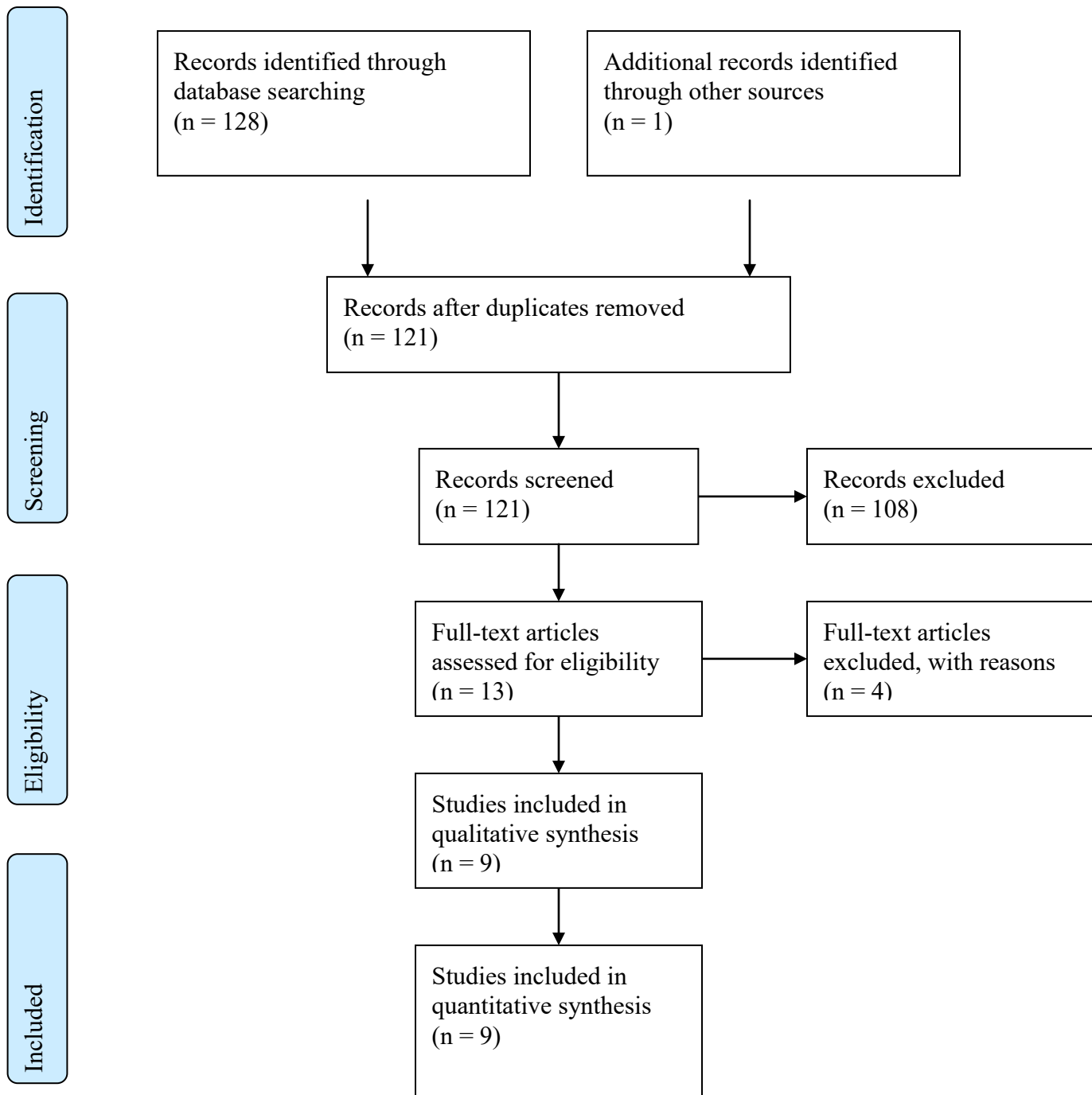
			Scoring based on zones around primary target.		Weeks and Kordus 1998.		4 – external-focused continuous (100%) or reduced (33%) feedback on movement pattern			for accuracy Effect size is not available.
Sidaway et al., 2012	48 TD children from grade 4 and 5 (31 boys); mean age 10.7 yrs	Pre-post observational study	72 trials (6 blocks x 12 trials) of throwing 100g cloth beanbags overhand to an unseen target placed 6 m away on	Compl ex	KR: positive numbers indicated overshooting and negative numbers	Visual	Group 1 & 3– task performed in standing or walking with continuous (100%) feedback	Absolute Error – at acquisition, retention and transfer phases Variable Error – standard deviation about the mean score	Retention and transfer phases (with children throwing the bean bag underhand) measured at 1 week follow-up	No difference between groups and task difficulty during acquisition. Better accuracy for Group 1 (simple task) and Group 4 (difficult task).

			the floor over a barrier. Performed under two conditions: standing and walking		indicated undershooting the target zone		Group 2 & 4 – task performed in standing or walking with reduced (33%) feedback			Similar results for transfer test. Effect size is not available.
Burtner et al., 2014	19 children with spastic CP aged 8-16yrs (9 boys); 20 TD	Randomized study - low-quality RCT	Motor task involving moving a lightweight lever 200 times (5 x 10 trial blocks for 4 sessions) of	Simple	KR: 1. Numerical total score; 2. RMSE; 3. Graphic representation	Visual	Group 1 & 3–Children with CP and TD: Continuous feedback (100%). Group 2 & 4 –	Performance accuracy (RMSE), consistency (Variable Error)	Retention test 24 hrs after acquisition	TD children: better accuracy and consistency at all phases compared to children with CP Group 1 & 3 (continuous

	children (12 boys) aged 8- 14 yrs		discrete horizontal elbow flexion/exte nsion movements		ta-tion of moveme nt super- imposed on target pattern		Children with CP and TD: Reduced feedback (62%); feedback gradually faded (100%, 75%, 50%, 25%).			feedback): fewer errors compared to Group 2& 4 (reduced feedback) in both acquisition and retention phases(effect sizer = 0.70*)..
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CP: Cerebral Palsy; TD: Typically-Developing;SCMAT: Score California Motor Accuracy Test; RCT: Randomized Controlled Trials;
 KP: Knowledge of Performance; KR: Knowledge of Results; RMSE: Root Mean Square Error, *effect size available from the cited
 publication, **effect size computed from publication data

Figure 4-1 Prisma Flowchart



CHAPTER 5 - VALIDATION OF REACHING IN A 2d VIRTUAL ENVIRONMENT IN TYPICALLY DEVELOPING CHILDREN AND IN CHILDREN WITH MILD HEMIPLEGIC CEREBRAL PALSY

5.1 Preface

Results of the first manuscript described in Chapter 3 indicated that children who have better sensation (proprioception and tactile threshold) were able to have better learning in comparison to children who had altered sensation. These results led to the question of whether the use of extrinsic feedback could act as a supplement to this reduced sensation.

In Chapter 4, the lack of evidence to identify which modalities of extrinsic of feedback that should be prioritized in TD children and in children with CP was identified. Factors that influenced the interpretation of the results included the lack of standardized intervention and assessments. Furthermore, none of the studies reported which features of the movement were learned and some studies were missing information regarding the parameters of the given feedback. In order to standardize and to manipulate the extrinsic feedback, it was recommended to use a VR system to deliver an intervention. However, as discussed in Section 2.7.3, perception in a VE in comparison to the physical environment may differ due to the quality of the viewing scene, the quality of the tracking system, the ongoing development and the impairments in children with CP (Tao et al., 2013; Ustinova et al., 2010). All these factors need to be considered as they affect the movement behavior and could ultimately results in undesired movement. Reinforcement of an undesired movement behavior such as a compensatory trunk movement may lead to maladaptive plasticity (Cirstea and Levin, 2000). Before the implementation of a VR system into an intervention, it is important to understand the possible limitations on reaching movements. Results of previous studies done in healthy adults and in individuals with stroke suggest slower movements and the increase use of shoulder and elbow range of motion. However, these results are not directly application to children. The first and second objective of this current study (Specific Objective 4 of Thesis) was to compare reaching kinematics made in a 2D VE to those made in a physical environment in TD children and in children with mild hemiplegic CP and the identify group and group by environment interactions. The third objective of this current study (Specific Objective 5 of Thesis) was to determine the relationships between

sensory impairments and reaching kinematics in children with CP. To do so, different reaching kinematics made in 3 planes in a 2D virtual environment were compared to those made in a matched physical environment in TD children and in children with CP. Comparison of the movements provided information on whether movements are affected by the quality of tracking and the VE.

Validation of reaching in a 2D virtual environment in typically developing children and in children with mild hemiplegic Cerebral Palsy

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Keywords: Children, kinematics, upper limb, virtual reality, cerebral palsy

This paper is currently under revision in Developmental Medicine & Child Neurology.

5.2 Abstract

Objectives: We compared three reaching movements made in two planes between 1) a low-cost game-based virtual reality (VE) and matched physical environment (PE); and (2) typically-developing (TD) children and children with Cerebral Palsy (CP).

Methods: Observational study in which children (TD, $n=17$, mean age 13.0 yrs; CP, $n=10$, 13.8 yrs) made 15 trials of each of three gestures (sagittal, frontal, vertical) in each environment. Upper-limb and trunk kinematics were recorded using an electromagnetic system (G4, Polhemus, 6 markers, 120Hz).

Results: Compared to PE, movements in VE made by TD children were slower ($p<0.002$), involved less trunk flexion ($p<0.002$) and rotation ($p<0.026$). In CP, trajectories were more curved ($p<0.005$) and children used less trunk flexion ($p<0.003$) and rotation ($p<0.005$). Elbow and shoulder kinematics differed from 2.8 to 155.4% between environments in both groups. Between groups, there were small, clinically insignificant differences with only the vertical gesture being longer in TD children. In children with CP greater tactile impairment was related to use of more trunk displacement.

Interpretation: Clinicians and researchers need to be aware of differences in movement variables when setting goals or designing protocols for improving reaching in children with CP using low-cost, game-based virtual reality systems.

5.3 Introduction

There is growing interest in using virtual reality (VR) technology to improve sensorimotor function in children with cerebral palsy (CP; Levac and Galvin, 2013). VR is a computer-based technology used to create virtual environments (VE) in which movement practice can be intensified and real-time interactions can occur (Schultheis et al., 2002). VEs can optimize neuroplasticity and learning by enhancing motivation and manipulating motor learning variables and task difficulty according to the user (Bryanton et al., 2006).

However, perception of object location in VE may differ from physical environments (PEs) due to limited or altered visual cues (e.g. display resolution, shadows, drop lines, perspective; Mon-Williams and Bingham, 2008). Self-motion perception and accuracy of movement-related feedback may be affected by decreased motion tracking accuracy, affecting motor learning (Tao et al., 2013). For example, reaching movements made in 2D video-capture and 3DVEs were slower and more curved than those in a matched PE in adults with and without stroke (Knaut et al., 2009; Liebermann et al., 2012). For 2D reaching, healthy adults used more elbow extension than for a matched task in PE due to uncertainty of target distance (Viau et al., 2004). Object location in VEs could be perceived differently by typically-developing (TD) children and children with mild hemiplegic CP compared to adults due to differences in previous experience (Newell and Verhoeven, 2017), memory and developmental stage (Green and Wilson, 2014). Sensory impairments may also affect learning in VEs in children with hemiplegic CP (Robert et al., 2013a).

Thus, comparing movements made in a low-cost, game-based VE with PE may determine whether movements are affected by the quality of motion tracking and the visual environment to inform clinicians about what treatment goals can be achieved using such systems.

Objective 1 was to compare upper-limb and trunk kinematics of three gestures made in two planes in a 2DVE to those made in a PE in TD children and children with CP. Objective 2 was to identify group and group by environment interactions. The third objective was to determine the relationships between sensory impairments and reaching kinematics in children with CP. Based

on previously identified differences in endpoint kinematics (Liebermann et al., 2012; Viau et al., 2004) for movements made in VEs, we hypothesized that 1) reaches made in the VE would be slower and more curved, and made with altered shoulder, elbow and trunk kinematics compared to those made in a matched PE in both TD children and children with CP; 2) kinematics would differ between groups and environments; and 3) altered kinematics in children with CP would be related to sensory deficits.

5.4 Methods

5.4.1 Participants

Out of 36 children contacted, 17 TD children (8 male; mean= 13.0±2.2yr; 14 right-handed) and 10 children with mild hemiplegic CP (9 male; mean= 13.8±1.8yr; 5 right-handed; Manual Ability Classification System score, MACS= I-II) were recruited. Reasons for non-participation included scheduling problems and non-compliance with inclusion criteria. Children were included if they could sit unsupported, reach in each direction, understand instructions, had full active range of motion, had functional vision and were cooperative. In addition, children with CP were included if they had motor impairment and spasticity in at least one arm. Exclusion criteria were presence of other neurological or musculoskeletal impairments or uncorrected vision. Parents and children signed informed consent/child-assent forms approved by the Centre for Interdisciplinary Research in Rehabilitation Ethics Committee.

5.4.2 Experimental Procedure

Children participated in one clinical/experimental session in this observational study.

5.4.2.1 Clinical Session

For sensory evaluation, tactile thresholds on the hand dorsum were assessed with valid and reliable Semmes-Weinstein filaments (S-W; Lafayette Instruments, USA). S-W values in 11-17 yr old TD children are 2.83 (Dua et al., 2016). Upper-limb proprioception (thumb, wrist, elbow, shoulder) was scored from 0 to 2 pts using the Fugl-Meyer scale, where 2 indicates no deficit, for

a maximal score of 8 (Fugl-Meyer et al., 1975). The Fugl-Meyer scale also assessed range of motion of 12 movements of fingers, wrist, forearm, elbow and shoulder on 3-point scales where 24 points represented full, painless range (Fugl-Meyer et al., 1975). Elbow flexor spasticity was assessed using the Tardieu scale (Tardieu et al., 1954), a 6-level ordinal scale classifying resistance felt by the examiner during passive stretching done at three different velocities. Manual Ability Classification System level (I to V; Eliasson et al., 2006) and handedness (Edinburgh Handedness Inventory; Oldfield, 1971) were also measured.

5.4.2.2 Experimental Session

Children sat comfortably on an adjustable chair with feet supported and their unrestricted trunk held close to the chair back (Fig. 5-1A). The arm was initially alongside the body with the elbow flexed to 90° and the hand over the navel. The contralateral arm was alongside the body. At an auditory signal, children performed a sagittal, frontal or vertical arm movement at a comfortable speed as accurately as possible. Since gestures did not require high-precision and differences between dominant and non-dominant arms were not expected, gestures were made by the dominant arm in TD children and the more-affected arm in children with CP.

5.4.2.3 Physical Environment

In PE, 7 round targets (65 mm radius) positioned on a wooden frame guided the gestures (Fig. 5-1B). Target 1 was in front of the participant at maximal arm length (measured from medial axilla to distal wrist crease with extended elbow; Schneiberg et al., 2010) plus 50 mm to avoid haptic feedback that could result in altered reaching kinematics (Levin et al., 2015). Children were explicitly instructed not to touch the targets to duplicate haptic conditions in both environments.

The game required children to trace 3 different trajectory paths to reach final targets and then to return their hand to the initial position. An anterior-posterior gesture was made in the sagittal plane in which children moved their hand through Targets 1, 4 and 5 and then returned it to the initial position via Targets 4 and 1. A medio-lateral gesture was made in the frontal plane, consisting of moving the hand through Targets 2, 1 and 3 and returning via Targets 1 and 2. A vertical gesture (i.e., up-down) was made in the sagittal plane in which children moved their

hand through Targets 7, 6, 1 and back via Targets 6 and 7. Fifteen trials were recorded per gesture but only movements of matched target distances were analyzed.

Participants practiced 5 trials per environment prior to recording for task familiarization. To avoid fatigue, 2 min rest periods were allowed between gestures. Gesture sequence was randomized using a permuted-block procedure to avoid learning. A successful reach occurred when the hand was within $\pm 5\text{cm}$ of the target center visually determined by the examiner and signaled verbally to the child.

5.4.2.4 Virtual Environment

A 2DVE with 3D rendering (Jintronix Rehabilitation System, Montreal) was displayed on a 29" screen. Arm and trunk movements were tracked with a Kinect camera (Microsoft, Redmond, USA) placed 1.5 m in front of the participant (Fig. 5-1C). The VE reproduced the 3 gestures (sagittal, frontal and vertical) with the same dimensions as the PE using a sequence of coloured targets. To compare gestures between environments, target locations and distances were measured in the PE with the Polhemus system (see below) and reproduced in the VE. In VE, the hand avatar was a fish controlled directly by hand movements. The VE provided the user with knowledge of results on task success (i.e., a "ding" sound for successful task) and terminal knowledge of performance on movement precision and speed as percentage scores.

5.4.3 Data Collection

Movements were recorded with a wireless electromagnetic tracking system (Polhemus G4, Vermont; 120 Hz) with 6 sensors placed on the index metacarpo-phalangeal (MCP) joint, mid-forearm, mid-arm, ipsilateral and contralateral acromions and mid-sternum. Positional (x,y,z) and rotational (pitch, roll, yaw) data were low-pass filtered (10 Hz) and used to reconstruct 3D angles (Biryukova et al., 2000) using quaternions and Euler angles. Computations accounted for sensor locations and anthropomorphic data (arm length, distances between sensors and joint centers) using custom software.

5.4.4 Data Analysis

Only reliable kinematics for describing reaching in similarly aged children with CP were used (Schneiberg et al., 2010). Since endpoint spatiotemporal parameters do not distinguish between movements accomplished with and without motor compensations (Levin et al., 2009), movement quality variables (i.e., joint rotations measured in body-centered coordinates; Schneiberg et al., 2010) were also assessed. Thus, we described movement at two levels: 1) endpoint performance (movement time, time-to-peak velocity, distance, trajectory straightness) and 2) movement quality (shoulder abduction/flexion, elbow extension, trunk flexion/rotation).

Although the task involved a sequence of two to four movement segments for different gestures, only one segment per gesture was analyzed. Segment onset/offsets were identified by changes in endpoint movement direction, defined as times when endpoint tangential velocity rose above or fell below 10% of peak velocity for at least 50ms. For the sagittal gesture, the segment between Targets 1 and 5 (anterior-posterior movement) was analyzed. For the frontal gesture (medio-lateral), the segment between Targets 2 and 3 was analyzed. For the vertical gesture (up-down), the segment analyzed was between Targets 7 and 1.

Movement time (s), time-to-peak velocity (s) and endpoint distance (cm) were calculated based on the 3D displacement of the MCP marker for each segment. Trajectory straightness was measured with the index of curvature (IC) as the ratio of actual endpoint path length to that of a straight line joining initial and final positions, where 1 indicates an ideal straight line (Atkeson and Hollerbach, 1985).

For shoulder movements, the arm outstretched laterally in line with the shoulder was defined as 0° of shoulder horizontal abduction and the arm positioned alongside the body was as 0° for shoulder flexion. Elbow extension was defined as 180° with the arm outstretched. For trunk flexion, the initial position was 0° with positive values indicating forward trunk pitch. Trunk rotation was computed from mid-sternal marker where initial position was 0° and right rotation was positive. All data were considered in the same frame of reference by inverting data from the

left arm to the coordinates of the right arm.

5.4.5 Statistical Analysis

Sample size calculation performed in G*Power (version 3.1.9.2) and MANOVA with parameters $\alpha=0.05$, $1-\beta=0.95$, Effect size=0.30, 2 groups, and 9 measurements resulted in a total recruitment of 20 children.

Data normality was verified with Levene's tests. For Objectives 1 and 2, three (sagittal, frontal and vertical) two-way MANOVAs measured interactions between environments and groups. In each analysis, 4 endpoint performance and 5 movement quality measures were dependent variables whereas environments and groups were independent variables. For the third objective, Pearson correlations were done between kinematic variables and clinical sensory assessments. Analyses were done with SPSS (version 20.0, SPSS Inc, Chicago) and minimal significance levels of $p<0.05$.

5.5 Results

Two children with CP had increased tactile thresholds and no children had proprioceptive deficits (Table 5-1). All children completed the sagittal gesture in each environment but data from only 10 TD children for frontal and vertical gestures were used. Data from 7 TD children were excluded since gesture lengths differed by $>6\text{cm}$ between environments whereas gesture lengths were equivalent between environments for the 10 children with CP.

Examples of typical endpoint trajectories from one TD child and one child with CP made in two environments are shown in Figure 5-2. Overall, trajectories were more curved in VE for all three gestures compared to PE in all children with CP while they were only more curved for the vertical gesture in VE in all TD children.

5.5.1 Movements made by TD children in VE compared to PE

Overall, regardless of the gesture, TD children made slower movements ($F_{1,9}=18.323$; $p=0.002$) and used less trunk flexion ($F_{1,9}=18.372$; $p=0.002$) and rotation ($F_{1,9}=7.022$; $p=0.026$) in VE. For example, time to peak velocity of sagittal gestures in VE was 60 ms (21.8%) longer than those in PE ($p=0.018$, Fig. 5-3A, supplementary Table 1) and movement times were prolonged by 364 ms (51.4%) and 207 ms (32.5%) for frontal ($p=0.001$, Fig. 5-3B) and vertical ($p=0.007$; Fig. 5-3C) gestures, respectively. Sagittal and frontal movements were made with 40.7% (-3.3° , $p=0.008$) and 50.6% (-4.3° , $p=0.007$) less trunk flexion and 40.5% (-1.7° , $p=0.015$), and 45.7% (-3.8° , $p=0.012$), less trunk rotation respectively. Vertical movements were 3.9% less straight (0.04, $p=0.002$) and involved 30° (155.4%) more shoulder abduction ($p=0.001$). All other variables were similar.

5.5.2 Movements Made by Children with CP in VE Compared to PE

For children with CP, movement time was not affected but trajectories were more curved ($F_{1,9}=13.757$; $p=0.005$), and less trunk movement was used (flexion: $F_{1,9}=16.911$; $p=0.003$; rotation: $F_{1,9}=13.232$; $p=0.005$) in VE. For example, movement time was shorter by 109 ms (-14.9%) for the sagittal gesture ($p=0.036$, Fig. 5-3A, supplementary Table 2). Sagittal, frontal and vertical gestures were more curved by 2.8% ($p=0.035$), 5.8% ($p=0.006$, Fig. 5-3B) and 6.7% ($p=0.007$, Fig. 5-3C), respectively. Sagittal and frontal gestures were made with 49.0% (-4.8° , $p=0.018$) and 39.4% (-2.8° , $p=0.001$) less trunk flexion as well as 44.4%, (-2.0° , $p=0.015$) and 39.7% (-2.3° , $p=0.025$) less trunk rotation, respectively. Frontal movement used 73.6% more shoulder flexion (11.7° , $p=0.025$). Vertical movements were slightly longer by 1.4 cm (3.1%, $p=0.001$) and involved 54.9% more shoulder abduction (14.7° , $p=0.005$) and 58.1% more elbow extension (12.5° , $p=0.043$). All other variables were similar.

5.5.3 Movements made by TD children compared to children with CP

There were small differences between groups for sagittal and frontal gestures. TD children made faster movements (by 97ms, $p=0.042$) for the sagittal gesture and used 29.7% more trunk

rotation (1.95° , $p=0.024$) for the frontal gesture. However, the vertical gesture was longer in both environments by a mean of 10.2cm (19.3%, $p=0.032$) and involved 24.4% more shoulder flexion (14.6° , $p=0.017$) in TD children while it was slightly more curved by 2.8% ($p=0.047$) in children with CP.

5.5.4 Differences in Kinematics Between TD Children and Children with CP

There were no group by environment interactions aside from a longer movement by 4.8cm in VE for the frontal gesture in TD children compared to no difference in the children with CP ($F_{1,39}=4.138$, $p=0.049$; Tables 5-2 and 5-3).

5.5.5 Relationship with Clinical Status of Children with CP

Endpoint performance variables were not related to tactile thresholds or proprioception. However, in VE, greater sensory impairment (higher S-W thresholds) was related to altered movement quality variables (frontal- less shoulder abduction: $r=-0.702$, $p=0.024$; more trunk flexion: $r=0.686$, $p=0.003$; vertical- more trunk flexion: $r=0.734$, $p=0.016$; sagittal- greater trunk rotation: $r=0.736$, $p=0.015$). In the PE, greater sensory impairment was related to greater trunk flexion for the sagittal gesture ($r = 0.641$, $p=0.046$). Two children with the highest tactile thresholds (S5 and S8, Table 5-1) used the most (5.4 to 20°) trunk flexion.

5.6 Discussion

To our knowledge, this is the first study to compare upper-limb kinematics in children reaching in a VE to those in a PE. Similar to previous studies in adults, movements in 2DVE were slower and involved less trunk movement (Tao et al., 2013).

Overall, frontal and vertical gestures were more influenced by the VE than sagittal ones in both groups partially supporting Hypothesis 1. Differences in kinematics could be related to altered perception of the user's hand and/or of the hand's interaction with the object in VE. To interact effectively with an object, visual cues (e.g., object size constancy, shadows, drop-lines, etc.) are

required to correctly identify object location and distance. A VE lacking such visual cues may lead to distorted perception due to misinterpretation of depth (Kenyon and Ellis, 2014). Hence, the hand avatar in VE should represent the actual hand location with respect to the target for a higher fidelity of distance representation (Kenyon and Afenya, 1995).

The use of a hand avatar may be responsible for the smaller trunk displacement used in both groups. It is likely that children realized that the hand avatar movement alone was sufficient for reaching the virtual target. This may have resulted in the use of greater shoulder ranges of motion in the VE. Similar results have been reported in adults with stroke making arm movements in a 3DVE viewed through a head-mounted display (Subramanian and Levin, 2011). Depth representation through drop-lines and shadows in 2DVE may have contributed to better object depth perception, the preservation of movement time (Fig. 5-3) for the sagittal target and the use of a larger range of shoulder motion.

For all gestures, movement time was longer in VE compared to PE. Several studies have reported large Kinect tracking errors when targets are located remotely and not directly aligned in front of the participant (Tao et al., 2013). These errors may lead to inaccurate estimates of real distance, which may be responsible for slower movements (Rochat and Wraga, 1997). The increased movement time in VE may also be explained by the perceptual uncertainty of object location resulting in movement corrections as the hand approaches the object (Loftus et al., 2004), or more curved trajectories.

For Hypothesis 2, differences in the use of shoulder flexion for the vertical gesture between groups can be explained by the longer distance moved by TD children in both environments. Children with CP may have under-reached the target by taking advantage of the Kinect tracking error reported to be as much as 8.7cm for the endpoint allowing children to successfully reach the virtual target using a smaller movement (Tao et al., 2013). Since determination of reaching distance in PE was based on the movements made in VE, the smaller distance would also apply to PE.

All other differences between groups were considered small and clinically insignificant, possibly

due to the mild nature of motor impairments in the children with CP, who are likely to have similar levels of activity compared to their healthy peers (Bult et al., 2013).

There was no relationship between sensory impairments, endpoint performance and most movement quality variables. Although slower movements in children with CP have been associated with decreased precision due to lack of feedback information, particularly proprioception (Bingham and Pagano, 1998), children in our study had intact proprioception and only mild tactile impairments. However, consistent with Hypothesis 3, increased trunk displacement for all gestures in VE and for the sagittal gesture in PE were related to the presence of tactile deficits. This may be related to the lack of availability of visual feedback for the correction of trunk movement in these children.

5.7 Limitations

Results of this study are limited by the small number of participants. Interpretation of the results cannot be extended to children with moderate-to-severe CP.

5.8 Conclusion

Small to large differences (2.8-155%) in reaching kinematics were observed for 3 gestures performed in a 2DVE compared to a matched PE in TD children and in children with CP. Aside from trajectory distance for the vertical movement, reaching kinematics were similar for all three gestures between groups. Children with greater tactile deficits used more trunk displacement.

Our results suggest that if low-cost, game-based VR systems such as the one studied here, are used as adjunctive therapies for upper-limb motor training in children with mild CP, differences in kinematics should be taken into consideration when setting training goals.

Future studies should investigate reaching kinematics in game-like VEs with better 3D rendering and movement tracking, compared to PEs. Studies in children with more severe cognitive and

motor impairments should also be done since these could impact movement strategies and perception in VEs.

5.9 References

References for this manuscript can be found at the end of the thesis in the Reference section.

Table 5-1: Demographic data and clinical parameters for children with cerebral palsy

Child	Age (y/m)	Gender	Side of hemiplegia	MACS	S-W	ROM /24	Proprioception /8
1	17.1	F	Right	1	2.83	24	8
2	13.0	M	Left	1	2.83	24	8
3	11.10	M	Right	1	2.83	24	8
4	14.9	M	Left	2	2.83	22	8
5	12.6	M	Right	2	4.31	18	8
6	15.4	M	Right	1	2.83	22	8
7	12.2	M	Left	2	2.36	23	8
8	16.2	M	Right	2	4.31	20	8
9	13.9	M	Left	2	3.22	24	8
10	13.3	M	Left	1	2.36	24	8

Abbreviations: F = female, M = Male, MACS = Manual Ability Classification System, S-W = Semmes-Weinstein, ROM = Range of motion. For S-W, the normal value is 2.83 (Dua et al., 2016).

Table 5-2 Mean (SD) kinematic data of sagittal, frontal and vertical gestures made in a physical (PE) and a virtual (VE) environment in typically-developing children. Absolute and percentage differences between PE and VE are indicated. Significant p values are shown in bold font.

Gestures	Parameters	PE	VE	Difference (%)	CI	F value	p value
Sagittal	Movement Time (s)	0.748 (0.190)	0.791 (0.168)	0.043 (5.7%)	0.683, 0.855	1.200	0.291
	Time to Peak Velocity (s)	0.275 (0.066)	0.335 (0.091)	0.060 (21.8%)	0.271, 0.340	7.060	0.018
	Trajectory Straightness (IC)	1.05 (0.04)	1.05 (0.05)	0.00 (0%)	1.03, 1.07	0.017	0.898
	Endpoint distance (cm)	45.5 (7.1)	44.7 (5.1)	-0.8 (-1.8%)	42.2, 48.0	0.260	0.617
	Shoulder Flexion (deg)	27.6 (9.4)	30.0 (9.8)	2.4 (8.7%)	23.1, 31.6	0.064	0.804
	Shoulder Abduction (deg)	58.7 (29.0)	66.1 (25.9)	7.4 (12.6%)	49.3, 75.4	1.435	0.250
	Elbow Extension (deg)	65.0 (22.0)	69.7 (24.1)	4.7 (7.2%)	57.0, 77.7	0.576	0.460
	Trunk Flexion (deg)	8.1 (4.1)	4.8 (2.8)	-3.3 (-40.7%)	4.9, 7.9	9.179	0.008
	Trunk Rotation (deg)	4.2 (2.3)	2.5 (1.1)	-1.7 (-40.5%)	2.6, 4.0	7.465	0.015
Frontal	Movement Time (s)	0.708 (0.186)	1.072 (0.200)	0.364 (51.4%)	0.776, 1.004	27.873	0.001
	Time to Peak Velocity (s)	0.383 (0.127)	0.405 (0.101)	0.022 (5.7%)	0.342, 0.445	0.153	0.705
	Trajectory Straightness (IC)	1.09 (0.15)	1.08 (0.04)	-0.01 (-0.9%)	1.02, 1.15	0.060	0.811
	Endpoint distance (cm)	49.2 (14.1)	50.6 (13.3)	1.4 (2.9%)	40.33, 59.6	2.981	0.118
	Shoulder Flexion (deg)	16.9 (9.7)	23.0 (14.5)	6.1 (36.1%)	12.2, 27.7	2.823	0.127
	Shoulder Abduction (deg)	59.0 (16.6)	75.1 (22.9)	16.1 (27.3%)	55.8, 78.4	4.290	0.068
	Elbow Extension (deg)	16.6 (11.7)	21.2 (9.1)	4.6 (27.7%)	13.3, 24.5	1.120	0.318
	Trunk Flexion (deg)	8.5 (3.9)	4.2 (0.9)	-4.3 (-50.6%)	4.9, 7.8	11.876	0.007
	Trunk Rotation (deg)	8.5 (3.7)	4.7 (2.4)	-3.8 (-45.7%)	4.8, 8.3	9.951	0.012
Vertical	Movement Time (s)	0.636 (0.130)	0.843 (0.139)	0.207 (32.5%)	0.670, 0.809	12.373	0.007
	Time to Peak Velocity (s)	0.359 (0.052)	0.346 (0.080)	-0.013 (-3.6%)	0.316, 0.389	0.193	0.671
	Trajectory Straightness (IC)	1.03 (0.03)	1.07 (0.04)	0.04 (3.9%)	1.03, 1.07	18.541	0.002
	Endpoint distance (cm)	56.0 (16.3)	56.1 (16.7)	0.1 (0.2%)	44.3, 67.8	0.090	0.771
	Shoulder Flexion (deg)	58.3 (20.6)	59.7 (22.8)	1.4 (2.4%)	44.7, 73.4	0.064	0.806
	Shoulder Abduction (deg)	19.3 (8.9)	49.3 (16.9)	30.0 (155.4%)	27.1, 41.4	27.143	0.001
	Elbow Extension (deg)	27.4 (11.6)	27.4 (11.7)	0.0 (0.0%)	19.9, 34.9	0.001	0.982
	Trunk Flexion (deg)	7.7 (2.1)	5.5 (2.9)	-2.2 (-28.6%)	5.7, 7.5	2.664	0.137

Trunk Rotation (deg)	3.4 (1.1)	3.0 (2.3)	-0.4 (-11.8%)	2.4, 4.0	0.220	0.650
CI = 95% confidence interval						

Table 5-3 Mean (SD) kinematic data of sagittal, frontal and vertical gestures made in a physical (PE) and a virtual (VE) environment in children with Cerebral Palsy. Absolute and percentage differences between PE and VE are indicated. Significant p values are shown in bold font.

Gestures	Parameters	PE	VE	Difference (%)	CI	F value	P value
Sagittal	Movement Time (s)	0.732 (0.129)	0.623 (0.167)	-0.109 (-14.9%)	0.583, 0.772	6.089	0.036
	Time to Peak Velocity (s)	0.287 (0.067)	0.265 (0.075)	-0.022 (-7.7%)	0.230, 0.323	1.430	0.262
	Trajectory Straightness (IC)	1.06 (0.03)	1.09 (0.07)	0.03 (2.8%)	1.04, 1.11	6.130	0.035
	Endpoint distance (cm)	43.5 (5.0)	44.5 (4.3)	1.0 (2.3%)	40.7, 47.3	4.202	0.071
	Shoulder Flexion (deg)	24.1 (7.4)	27.2 (10.2)	3.1 (12.9%)	20.9, 30.5	0.652	0.440
	Shoulder Abduction (deg)	59.7 (23.2)	51.4 (28.4)	-8.3 (-13.9%)	38.0, 73.1	2.644	0.138
	Elbow Extension (deg)	62.9 (17.1)	52.0 (19.9)	-10.9 (-17.3%)	47.2, 67.8	3.612	0.090
	Trunk Flexion (deg)	9.8 (4.7)	5.0 (3.9)	-4.8 (-49.0%)	4.9, 9.9	8.403	0.018
	Trunk Rotation (deg)	4.5 (2.4)	2.5 (1.3)	-2.0 (-44.4%)	2.4, 4.7	9.019	0.015
Frontal	Movement Time (s)	0.757 (0.294)	0.811 (0.265)	0.054 (7.1%)	0.647, 0.922	0.177	0.684
	Time to Peak Velocity (s)	0.371 (0.109)	0.400 (0.123)	0.029 (7.8%)	0.334, 0.437	0.259	0.623
	Trajectory Straightness (IC)	1.04 (0.02)	1.10 (0.06)	0.06 (5.8%)	1.05, 1.10	12.902	0.006
	Endpoint distance (cm)	44.8 (7.1)	45.3 (7.1)	0.5 (1.1%)	40.1, 50.0	0.670	0.434
	Shoulder Flexion (deg)	15.9 (6.6)	27.6 (12.0)	11.7 (73.6%)	16.9, 26.3	7.220	0.025
	Shoulder Abduction (deg)	52.7 (14.0)	62.5 (19.8)	9.8 (18.6%)	49.1, 66.2	1.589	0.239
	Elbow Extension (deg)	23.2 (12.1)	27.3 (15.0)	4.1 (17.7%)	18.8, 31.7	0.387	0.549
	Trunk Flexion (deg)	7.1 (2.2)	4.3 (1.9)	-2.8 (-39.4%)	4.4, 7.0	24.025	0.001
	Trunk Rotation (deg)	5.8 (2.5)	3.5 (1.2)	-2.3 (-39.7%)	3.6, 5.6	7.163	0.025
Vertical	Movement Time (s)	0.728 (0.262)	0.700 (0.210)	-0.028 (-3.9%)	0.560, 0.868	0.192	0.672
	Time to Peak Velocity (s)	0.369 (0.123)	0.324 (0.102)	-0.045 (-12.2%)	0.270, 0.423	4.017	0.076
	Trajectory Straightness (IC)	1.05 (0.02)	1.12 (0.07)	0.07 (6.7%)	1.05, 1.11	12.330	0.007
	Endpoint distance (cm)	45.1 (7.4)	46.5 (7.7)	1.4 (3.1%)	40.4, 51.2	23.759	0.001
	Shoulder Flexion (deg)	40.1 (13.3)	48.7 (15.4)	8.6 (21.5%)	38.3, 50.5	1.401	0.267
	Shoulder Abduction (deg)	26.8 (11.3)	41.5 (20.3)	14.7 (54.9%)	23.3, 45.0	13.534	0.005
	Elbow Extension (deg)	21.5 (11.1)	34.0 (14.7)	12.5 (58.1%)	20.7, 34.9	5.540	0.043
	Trunk Flexion (deg)	6.4(4.7)	4.7(2.2)	-1.7 (-26.6%)	3.5, 7.6	1.556	0.244
	Trunk Rotation (deg)	3.6 (2.4)	2.4 (0.7)	-1.2 (-33.3%)	2.1, 3.8	2.222	0.170

CI = 95% confidence interval

Figure 5-1 The experimental setup and the three gestures in the two environments.

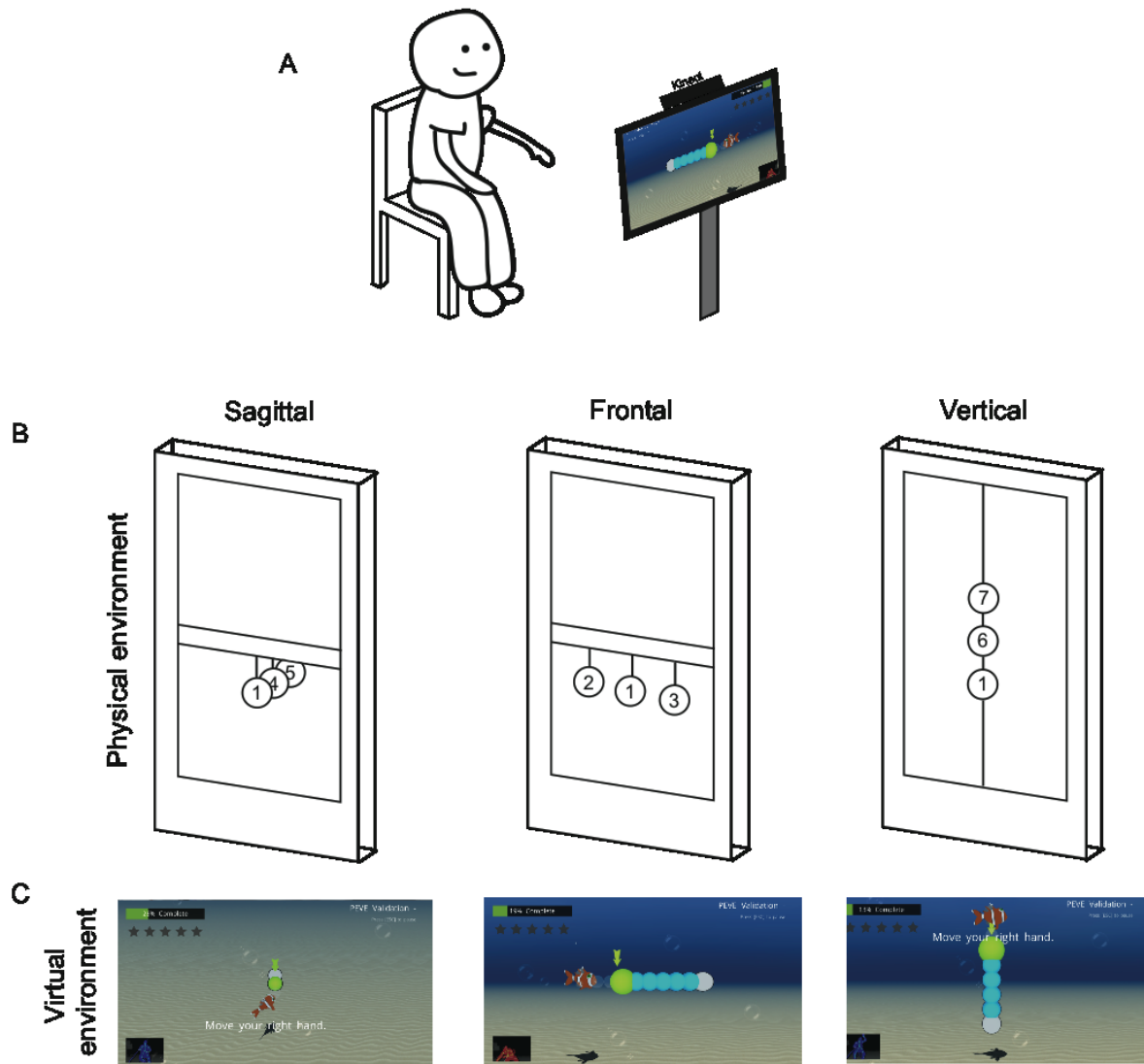


Figure 5-2 Typical endpoint trajectories of sagittal, frontal and vertical movements made in both physical and virtual environments in TD children and in children with CP.

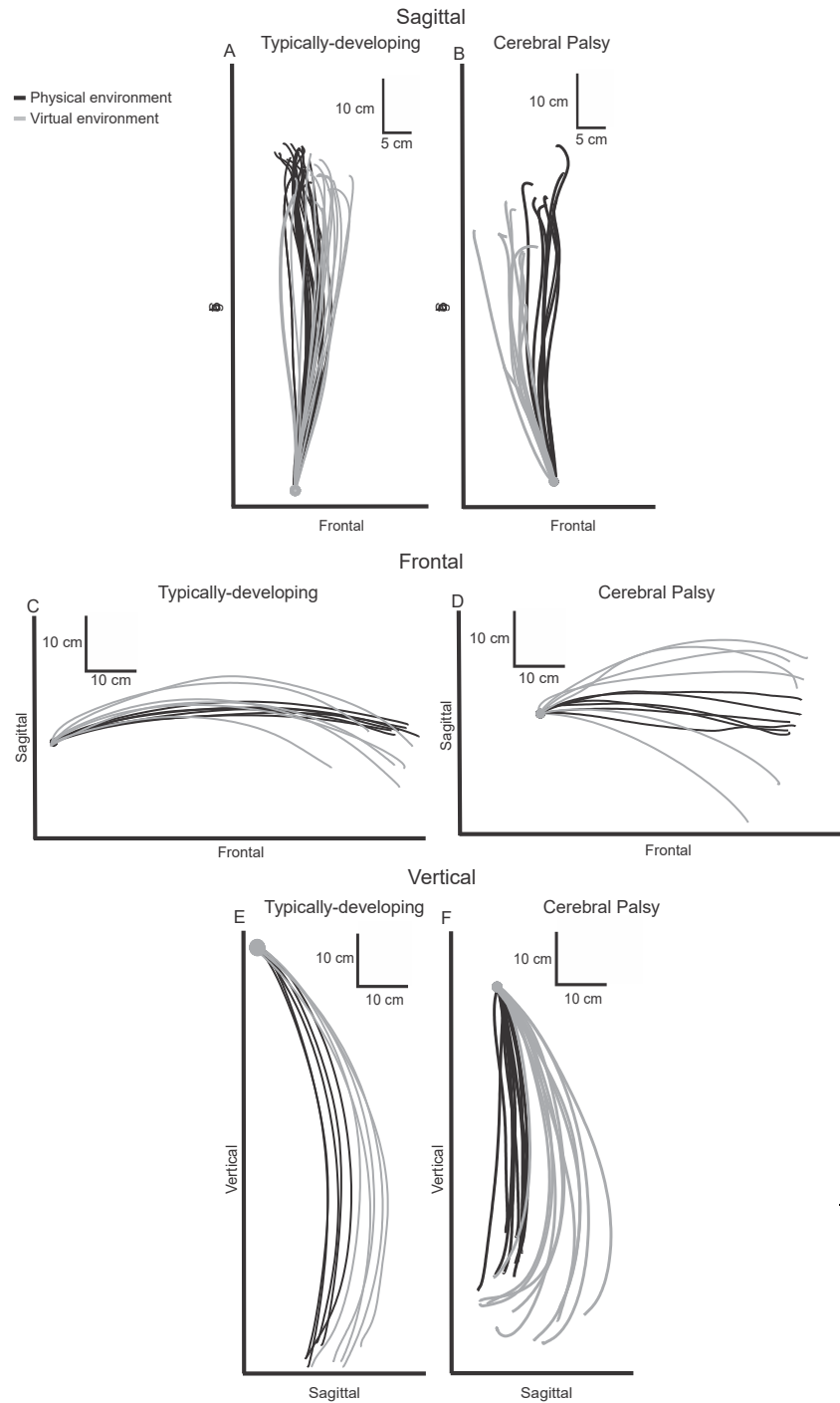
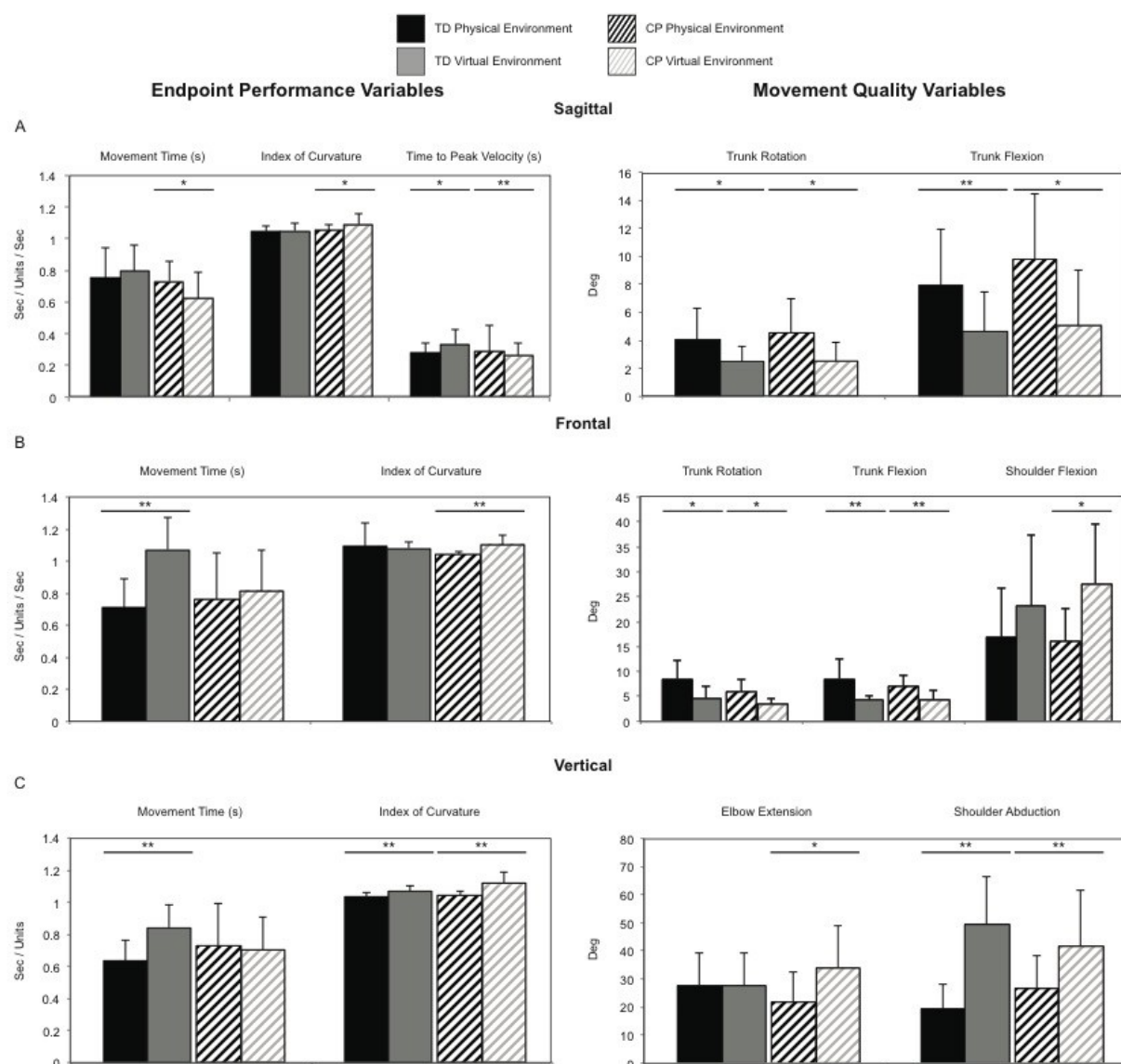


Figure 5-3 Histograms of endpoint performance and movement quality variables.



CHAPTER 6 - FEASIBILITY OF DELIVERING FEEDBACK IN A VIRTUAL REALITY SYSTEM FOR CHILDREN WITH CEREBRAL PALSY

6.1 Preface

The second study presented indicated the importance to identify the most suitable types and frequencies of extrinsic feedback to optimize motor learning. The results from the systematic review were not conclusive as which modalities and frequencies of feedback that needs to be prioritized to optimize upper limb motor learning in children with CP. One possible way to study the different modalities of feedback is through the use of VR. As discussed in Section 2.2, VR has advantages, such as the provision of high motivation and more importantly for the objective of this thesis, the possibility to manipulate and to standardize the delivery of feedback. However, the possible alteration of perception within a virtual environment is to be considered as it could influence the movement. Hence, results of the third study (see Chapter 5) indicated that there are some limitations that a clinician or a researcher needs to be aware when using a VR system to deliver an intervention. Particularly, the results of the third manuscript indicated that movements tend to be slower and more curved while using less trunk flexion and rotation in the VE. These differences in movements need to be accounted for when it comes to develop a research paradigm using a VR system as it may possibly affect the movement behavior.

The objectives of the fourth manuscript was to determine the feasibility of delivering different frequencies (faded vs continuous) of extrinsic feedback through a VR system for upper limb skill acquisition in children with mild hemiplegic CP and to determine the effectiveness of a short intensive training program with feedback delivered in VR. To do so, 7 children with mild hemiplegic CP participated in a four 2-hour training sessions using a low-cost game-based VR system in which the delivery of feedback was manipulated. Feasibility of this study was assessed using the attendance rate and participation satisfaction. Improvement and retention of upper limb motor skills were assessed with two clinical measures (Jebsen-Taylor Test and Melbourne Assessment) and with movement kinematics during a standardized reach-to-grasp task of an object at pre, post and one month following the intervention.

Feasibility of delivering feedback in a virtual reality system for children with Cerebral Palsy

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Keywords: Motor learning, feedback, children, cerebral palsy. kinematics, upper limb, virtual reality

This paper will be submitted in August, 2017.

6.2 Abstract

Background: The ability to improve and acquire new motor skills depends on motor learning defined as a set of processes associated with practice or experience leading to permanent change of movement performance. One way to optimize retention of motor skills is to provide extrinsic feedback to the learner. With the increased use of virtual reality (VR) systems in rehabilitation, it is now possible to deliver standardized extrinsic feedback. However, there is no evidence on the success of delivering standardized extrinsic feedback through a VR system in children with hemiplegic Cerebral Palsy (CP).

Objective: We determined 1) the feasibility of delivering different frequencies (faded vs continuous) of extrinsic feedback through a VR system; and (2) the feasibility to implement a short intensive VR intervention in children with mild hemiplegic CP.

Methods: Seven children with mild hemiplegic CP (mean age 13 yrs $\pm$ 1.2yr) participated in 4 2-hour training sessions using a low-cost, game-based VR system in which the delivery of feedback was manipulated. The frequency of additional numerical feedback on knowledge of performance (trunk displacement) and knowledge of results (accuracy and velocity of the hand movement) was either continuous (after every trial) or faded (reduced over time). Feasibility of this intervention study was assessed with the attendance rate and participant satisfaction. Arm and hand function was assessed using two clinical scales (Jebsen-Taylor Test and Melbourne Assessment) and with movement kinematics during a standardized reach-to-grasp task of an object placed at 3 distances (2/3, full and 1^{2/3} arm length) at pre, post and one month following the intervention.

Results: All participants completed all measures and reported moderate to high motivation. The delivery of different frequencies (continuous and faded) was feasible. All children improved or maintained Melbourne Assessment scores at post and follow-up assessments. Six children completed the Jebsen-Taylor Test faster at follow-up. Significant kinematic changes were obtained in all children. A minimum of 4 children either maintained or improved performance outcomes (movement time and trajectory straightness) for each distance. Movement quality

improved in a minimum of 2 children as demonstrated by the greater use of elbow extension and shoulder flexion for each distance. More than half of the children maintained or reduced their trunk flexion for reaches at each distance at post and follow-up.

Interpretation: This pilot study demonstrates the feasibility of implementing a VR intervention in which extrinsic feedback may be standardized. Delivery of continuous and faded extrinsic feedback using VR may optimize motor learning in children with mild hemiplegic CP but further studies are required.

6.3 Introduction

During an intervention, clinicians provide additional extrinsic feedback to their patients, with the objective to improve motor skills, which ultimately increases functional activities and participation. The ability to improve motor skills depends on motor learning, a concept defined as a set of processes associated with practice or experience leading to a relatively permanent change in the capability for movement (Schmidt and Lee, 2011). Optimization of motor learning can occur through the integration of multiple sources of information such as intrinsic feedback (e.g. proprioception, tactile threshold) and extrinsic feedback (visual, auditory), a concept defined as multisensory integration (Lickliter, 2011). Evidence from neurophysiological research indicates that sensory systems are linked together very early in development and that the brain is organized in a way to facilitate the integration of information across sensory modalities, enhancing a child's perception to have a proper response (Calvert and Thesen, 2004; Gori et al., 2011).

However, the provision of additional feedback could be impeded or used differently in children with cerebral palsy (CP) in comparison to their healthy peers due to impairments (e.g. reduced sensation, increase of motor disorders; (Robert et al., 2013) of the different physiological and neurological systems (Schmidt and Lee, 2011). With a prevalence of 1.5 to 2.5 per 1000 births, CP is the most common neurological disorder making it the most costly for the health care system (Johnston and Hoon, 2006; Paneth et al., 2006). A better understanding of the impact of providing additional feedback in rehabilitation to children with CP could optimize motor learning. Based on a recent literature review, the modalities of extrinsic feedback that need to be prioritized to optimize motor learning in typically-developing children and in children with CP remain elusive as there is limited evidence and lack of standardized interventions (Robert et al., 2017). For example, the type of extrinsic feedback that should be given remains unclear although it was suggested that a combination of both auditory and visual feedback should be prioritized, consistent with the notion of multisensory integration (Sigrist et al., 2013). The optimal delivery frequency of feedback is also undetermined. For instance, faded feedback (i.e., feedback reduced over time) may be more effective compared to continuous feedback (i.e., feedback provided after every trial) in adults (Subramanian et al., 2010), but there is limited evidence for the pediatric population, especially during a complex task (Robert et al., 2017). As for the feedback

parameters, there is no consensus on whether or not to provide feedback on the performance (knowledge of performance; KP) or the result of the task (knowledge of results; KR).

In rehabilitation, there is a growing interest in the use of low-cost, game-based virtual reality (VR) to improve motor function in children with CP (Levac and Galvin, 2013). VR is a computer-based technology that can be used to create virtual environments in order to enhance motivation, to manipulate principles of motor learning and to modify tasks difficulty according to user abilities and needs (Bryanton et al., 2006). It also provides the advantage of delivering real-time standardized extrinsic feedback using a combination of different modalities on either movement performance or movement quality that may promote adaptive neuroplasticity (Schultheis et al., 2002).

Consequently, the use of VR as an intervention could be implemented to deliver different frequencies of extrinsic feedback to optimize motor learning in children with CP. The main objective was to determine the feasibility (*i.e. implementation, acceptability*) of delivering extrinsic feedback through a VR system for upper limb skill acquisition in children with mild hemiplegic CP. We hypothesized that the delivery of different frequencies (continuous or faded) of extrinsic feedback using a VR system will be feasible. Our second objective was to determine the feasibility to implement a short intensive VR intervention in children with mild hemiplegic CP. We hypothesized that it will be feasible to implement a short intensive VR intervention in children with mild hemiplegic CP and that children will be improve either clinical or kinematic outcomes.

6.4 Methodology

6.4.1 Participants

Seven children with mild hemiplegic CP aged between 11 and 15 years old (7 male; mean age = 13 ± 1.2 yr) were recruited from two Quebec pediatric centers. Children were included if they could sit unsupported, were able to reach in any direction, understood instructions, had functional vision and were cooperative. In addition, children with CP were included if they were impaired in at least one arm and had spasticity. Participants were excluded if they had other neurological or musculoskeletal impairments or uncorrected vision. Participants were excluded if

they had any other neurological or musculoskeletal impairments or uncorrected vision, medical or surgical procedures in the 6 months leading up to or planned during the study period, or were receiving upper limb occupational or physical therapy. Cognitive deficits was not considered for this study. Parents and children signed informed consent/child-assent forms approved by the Centre for Interdisciplinary Research in Rehabilitation Ethics Committee.

6.4.2 Study Design

This study was a pre-post-follow up intervention paradigm (Fig. 6-1A) in children with mild hemiplegic CP. Children were randomly allocated to one of two feedback groups by an individual uninvolved with recruitment, evaluation or training. The two groups practiced repetitive reaching of four different activities that required different combinations of joint rotations in a VR training environment. The frequency of feedback provided by the VR system differed for each of the two groups (continuous, faded). The Jintronix software had to be programmed before the intervention to deliver either continuous or extrinsic feedback. In the continuous feedback group, numerical terminal feedback was given after each trial. In the faded feedback group, the frequency of numerical terminal feedback for the first session was continuous (feedback after every trial). For the next session, the frequency of feedback was reduced to 75% (i.e. the participant receive no feedback every 4th trial). In the third session, feedback frequency was reduced to 50% (feedback provided after every second trial). In the last session, feedback frequency was reduced to 25% (once every 4 trials).

Clinical and kinematic evaluations were conducted before (pre), immediately after (post) and one month following (follow-up) a 4 session intervention over a 10 day period (for a total of 7 sessions). Each intervention session consisted of 2-hours of practice (300 trials per session for a total of 1200 trials). A single evaluator, blinded to group assignment, completed all clinical outcome assessments. A kinesiologist blinded to the clinical assessment supervised the intervention and did the kinematic evaluation.

6.4.3 Intervention

For each training session, the participants were comfortably seated on an adjustable chair with the feet supported on the floor. Standardized instructions were given to the participants as to how to execute the required movement with minimal trunk displacement and at a comfortable speed.

The 2D virtual environment with 3D rendering called the Jintronix Rehabilitation System was developed by Jintronix Inc., Montreal. The virtual environment tracked arm and trunk movements with a Kinect camera (Microsoft, Redmond, USA) placed in front of the participant. Limitations of the tracking quality are discussed elsewhere (Tao et al., 2013). The virtual environment scene was displayed on a 29" screen and had 4 distinct interactive games (Fig. 6-1B) of which 2 involved unilateral arm movement (Fish Frenzy and Pixel Waves) and 2 involved bilateral arm movement (Catch, Carry and Drop, Pop Clap). Unilateral games required the child to trace different trajectory paths or reach a target with his arm whereas bilateral games involved clapping or catching an object with both hands. During sessions 1 through 3, two games were played (150 trials per game). In the last session, all 4 games were played (75 trials per game). Sessions were divided in 6 blocks (50 trials per block). A 2-minute rest period between blocks and games was given to the participants to avoid fatigue. Difficulty settings (speed and precision) of each game were determined for each individual according to his ability level, which was considered moderately difficult (8 successful movements out of 10 increased the game difficulty).

6.4.4 Augmented Feedback

Provision of concurrent feedback on KP (trunk displacement) and KR (time remaining to complete the task) were visually displayed on the screen for each game. Manipulated extrinsic feedback (continuous or faded) provided to the participants at the end of a trial was a combination of visual numerical KP (percentage loss because of excessive use of trunk displacement) and KR (movement precision and velocity).

6.4.5 Evaluations

6.4.5.1 Feasibility Outcomes

Two out of eight general areas of focus recommended for feasibility studies were assessed: attendance rate (implementation criteria) and participant satisfaction (acceptability criteria) (Bowen et al., 2009).

Implementation, defined as how an intervention can be fully applied as planned and proposed, was assessed by measuring the adherence to the training and evaluations, for a total of 7 sessions. The analysis of clinical and kinematic outcomes were also considered in the implementation criteria. Recruitment rate was defined as the number of children with CP contacted from those eligible.

Acceptability, defined as how the individuals react to the intervention, was measured via the valid intrinsic motivation inventory (IMI). The full version of the IMI consists of 45 multidimensional questions that assess the subjective experience of the participants related to the activities performed. The reliable version that was used consists of fifteen items, which assess the interest/enjoyment, perceived competence, effort/impression, value/usefulness (McAuley et al., 1989). Each item rated the statement in a range between 1 (not at all true) and 7 (very true) where 7 indicated high motivation.

6.4.5.2 Clinical Session

Clinical measures included the handedness of the participants using the valid and reliable 10-item Edinburgh Handedness Inventory (Oldfield, 1971) and the Manual Ability Classification System (Eliasson et al., 2006). Tactile threshold was measured at the index fingertip using the valid and reliable Semmes-Weinstein monofilaments (S-W; Lafayette Instruments, Lafayette, IN, USA). S-W values in typically developing children aged 5 to 9 years range from 2.83 to 3.61 mm (Auld et al., 2011). Upper limb range of motion of five arm joints and proprioception were

measured on numeric scales (Fugl-Meyer et al., 1975) with a maximal score of 24 and 8 respectively, indicating no impairment. Spasticity was measured in the elbow flexor muscles using the six-level ordinal Tardieu scale (Tardieu et al., 1954). A level 0 indicates no resistance throughout the passive movement whereas a level of 5 indicates the joint is immovable. The test is also performed at 3 different velocities where V1 is as slow as possible and V3 is as fast as possible. Upper limb impairment and function were measured with the valid and reliable Melbourne Assessment (Hoare et al., 2011; Randall et al., 2001). The Melbourne Assessment, which was videotaped and independently analyzed, assesses 16 arm and hand movements for range of motion, accuracy, fluency, quality, accomplishment and/or speed on 4- or 5-points scales for a maximal score of 122 points. Unimanual activities of daily living were assessed using the valid and reliable standardized timed Jebsen-Taylor Test (Jebsen et al., 1969; Stern, 1992; Taylor et al., 1973). The Jebsen-Taylor Test consists of 7 subtests including moving heavy and light objects, simulated feeding, stacking checkers, picking up small common objects and card turning.

6.4.5.3 Experimental Session

Arm and trunk movements during a standardized reach-to-grasp task to objects placed at three distances were recorded (Fig. 6-1C). This standardized task is widely used since it simulates a daily functional activity (Schneiberg et al., 2010; Schneiberg et al., 2002). Each participant was comfortably seated on an adjustable chair with the feet supported on the floor. In the initial position, participants were asked to keep his/her trunk close to the chair back support but trunk movement was not restricted during testing. The arm was initially positioned alongside the body with the elbow flexed to 90° and the hand over the navel. The contralateral arm was alongside the body. Upon an auditory signal, the participant performed the reaching task at a comfortable speed as accurately as possible to simulate bringing the object to the mouth. Although only the reach-to-grasp part of the reaching movement was analyzed, the whole reaching task was performed in order to ensure the task was ecologically valid. Seated children reached to grasp a 2 cm block placed at three distances proportional to the child's arm length (close target = 2/3 of the maximal arm length, full target = maximal arm length and far target = 1 + 2/3 of the arm length). Arm length was measured from the medial axillary border to the distal wrist crease with the

elbow extended (Schneiberg et al., 2010). Reaches to the far target tested the ability of the child to reach beyond arm length, a movement that was not practiced during the training.

Participants were allowed to practice two trials prior to recording to become familiar with each task. To avoid fatigue, participants had two-minute rest periods after each block of 15 trials (for a total of 45 trials). Each block represented one distance (close, full and far targets). The sequence of distances was randomized between children and sessions using a permuted-block randomization procedure.

6.4.6 Data Collection

Movements were recorded with a wireless electromagnetic tracking system (Polhemus G4, Vermont) and sampled at 120 Hz. Six sensors were placed at the following locations: metacarpophalangeal (MCP) joint of the index finger, mid-forearm, mid upper arm, ipsilateral and contralateral acromio-humeral joints and mid-sternum. Positional (x, y, z) and rotational data (pitch, roll, yaw) were low-pass filtered (10 Hz) and used to reconstruct 3D angles (Biryukova et al., 2000) based on sensor locations and anthropomorphic data using custom Matlab programs.

6.4.7 Data Analysis

Kinematic outcomes were restricted to those shown to be reliable for describing arm movements in similarly aged children with CP (Schneiberg et al., 2010). Two movement performance (movement time and trajectory straightness) and three movement quality (elbow extension, shoulder flexion, trunk flexion) measures were obtained. Segment onset/offsets were identified based on changes in endpoint movement direction. These points were defined as times at which endpoint (MCP marker) tangential velocity rose above or fell below 10% of the peak velocity for at least 50 ms.

Movement time (s) was calculated based on the 3D displacement of the MCP marker. Trajectory straightness was measured using the index of curvature (IC) as the ratio of actual endpoint path

length to the length of a straight line joining initial and final positions. An ideal straight line has an index of curvature of 1, while a semicircle has an index of curvature of 1.57 (Atkeson and Hollerbach, 1985). Shoulder flexion with the arm positioned alongside the body was defined as 0°. Elbow extension was defined as 180° in fully stretched position. Trunk flexion was calculated as the sagittal displacement of the mid-sternal marker, where the initial position was defined as 0° and forward trunk displacement was positive.

6.4.8 Statistical Analysis

A single-participant research analysis was used because of the small sample size and participant heterogeneity. To determine whether changes occurred in each parameter for each child at post-test and follow-up for each target, a regression with visual trend analysis and computed effect size was used. A linear regression line was fit through all the pre-test (baseline) data points and a horizontal straight line was extended from the end of the regression through post-test and follow-up data. The number of data points above or below the line was counted for each phase. Between-mean differences of post-test and pre-test values were divided by the pre-test standard deviation to determine the effect size. The same was carried out to calculate effect sizes at follow-up with post-test replaced by follow-up. Effect sizes of 0.20, 0.50 and 0.80 were considered small, moderate and large respectively (Portney and Watkins, 2009). Effect sizes greater than 0.50 identified motor learning and transfer based on previous studies (Cohen, 1988). Motor learning was defined by retention at follow-up of improvements in performance and or movement quality variables for the close target reach-to-grasp task and full target reach-to-grasp task. Transfer was defined as improvements in performance and/or movement quality variables at post-test for the far target reach-to-grasp task.

6.5 Results

Participant recruitment is outlined in the flow diagram (Fig. 6-2). All participants (7 males) with mild spastic hemiplegia (MACS levels I-II) were able to complete each phase of the study. All children except one had normal tactile threshold values and normal range of motion. Demographic characteristics and clinical scores for all participants are listed in Table 6-1.

As the number of participants for this study was considered low, the data were combined for all children.

6.5.1 Feasibility Outcomes

6.5.1.1 Implementation

Between March 2015 and January 2016, 49 children with CP were assessed for eligibility, 19 (39%) were eligible, and 7 were recruited (37%; Fig. 6-2). Reasons for non-participation included scheduling problems, trust issues with research, location of the research too far from home and financial constraints. Measurement completion rates were 100% for all measures.

6.5.1.2 Acceptability

All 7 children reported moderate to high motivation (4.6 to 6.47 on the IMI scale) during the intervention as demonstrated by the relatively high score (mean motivation = 5.45 ± 0.67). Hence, the acceptability of this intervention was considered high.

6.5.2 Clinical Outcomes

Five children maintained Melbourne Assessment score at post and follow-up evaluations. The other two children improved Melbourne Assessment score at both post and follow-up evaluations. Improvements in Jebsen-Taylor Test were observed in 5 children at post-test. Six children were faster to complete all 7 subtests on the Jebsen-Taylor Test at follow-up in comparison to the pre-test.

6.5.3 Performance Outcomes

Figure 6-3 shows examples of trend line analysis for the five kinematic outcomes at far target for child number one in the continuous group. No difference was observed on the movement time outcome. Trajectory was less curved at follow-up ($ES = -1.45$). Elbow extension was decreased

at post-test (ES = -0.72) but remained stable at follow-up. Range of shoulder flexion was decreased at post-test (ES = -0.67) and at follow-up (ES = -0.59). Less trunk flexion was used at post-test (ES = -1.13) but an increased use of trunk flexion was observed at follow-up (ES = 2.61).

6.5.3.1 Movement Time

Movement time was shorter by -0.065s (12%) in one child to the close target following the intervention (Table 6-2). At follow-up, movement time was shorter ranging from -0.086s (13.5%) to -0.098s (10%) in 4 children. For movements to the full target, one child improved by -0.100s (10.3%) whereas two children (-0.066s, 9.5%; -0.137s, 21.3%) made faster movements at follow-up. Transfer of improvements to the far target at post-test occurred in 2 children (-0.121s, 11%; -0.411s, 26.8%). Movement time was shorter by -0.220s (14.3%) in one child at follow-up.

6.5.3.2 Index of Curvature

For the close target, trajectories were 4.8% to 22.7% less curved in 3 children after the intervention (Table 6-3). Trajectories were less curved by 5.2% to 14.1% at follow-up in 3 other children. For the full target, 2 children made less curved movement by 11% and 12.7% whereas 3 children improved trajectory straightness (8.7% to 13.7%) at follow-up. For far target, 2 children made less curved movement by 6.3% and 12.6% in 2 children at post-test. Transfer of improvements at follow-up occurred in 5 children (3.1% to 10.8%).

6.5.4 Movement Quality Outcomes

6.5.4.1 Elbow Extension

For the close target, elbow extension range increased by 35% (14.7°) and 18.1% (18.1%) in 2 children at post-test (Table 6-4). One child used 19.4% (6.3°) more elbow extension for the close target at follow-up. For the full target, 2 children improved elbow extension (6.7°, 13.3%; 19°, 51.3%) and retained the improvement (4.6°, 9.2%; 24.1°, 64.9%) a month after the

intervention. Transfer of improvements to the far target at post-test involved the use of 63% (27.7°) more elbow extension in 1 child and ranged from 13.2% (6.2°) to 55% (24°) in 3 children at follow-up.

6.5.4.2 Shoulder Flexion

None of the children increased the use of shoulder flexion for the close target (Table 6-5). However, 2 children used more shoulder flexion (8.6°, 15.6%; 11.8°, 35%) at follow-up. Shoulder flexion increased by 46.4% (6.5°) and 32.2% (6.7°) for reaches to the full target in 2 children. Improvements of shoulder flexion were observed in 2 children at follow-up (7.2°, 51.1%; 7.4°, 39.2%). Improvement of shoulder flexion was transferred at the post-test in 2 children (14.3°, 61.5%; 8.0°, 24.4%). Retention of the improvement was observed in 2 children (20.5°, 89%; 5.9°, 18%).

6.5.4.3 Trunk Flexion

Decreased use of trunk flexion ranging from 44.4% (-1.1°) to 72.1% (-2.2°) in 3 children was observed for reaches to the close target but no child retained the improvement at follow-up (Table 6-6). For the full target, less trunk flexion (-5.1°, 54.6% to -16.3°, 74%) was observed in 3 children at post and follow-up. Less trunk flexion (-7.6°, 51.2% to -24.7°, 39%) was observed in 6 children for the far target at post-test. Reaches to the far target at follow-up involved 53.3% (-34.1°) less trunk flexion in 1 child.

6.6 Discussion

Our results indicate the feasibility of delivering extrinsic feedback through a low-cost, game-based VR intervention. Moreover, it is feasible to deliver either continuous or faded feedback using a VR system in children with mild hemiplegic CP. Previous studies reported the importance of developing more robust research designs in which different modalities of extrinsic feedback could be compared (Robert et al., 2017; Sigrist et al., 2013; Subramanian et

al., 2010). Our results also support the possibility that children with mild hemiplegic CP could improve upper limb skills following an intensive VR intervention.

The number of participants eligible for this study was considered low to moderate with only a third of them agreeing to participate. Recruitment was limited due to several factors such as financial and time restraints. Implementation of this VR intervention in a setting in which recruitment is less of a challenge and which has better access and resources could potentially increase the number of participants. For example, such an intervention design could be integrated into summer camps designed for children with CP (Ballaz et al., 2011; Kuo et al., 2016). These camps usually have the resources (financially and personnel) to provide an intensive camp in which factors influencing motor learning could be optimized (e.g. intensity).

Measurement completion rate for this intervention was excellent. The research paradigm in our facility was well-received by the participants as reported by the high level of motivation and by the positive attitudes of the families. This, coupled with the moderate success of implementation indicates the feasibility of delivering a VR intervention in which different frequencies of feedback could be manipulated and standardized. Other areas of focus of a feasibility study such as the demand, the integration and expansion of the intervention were not assessed (Bowen et al., 2009).

Changes of the individual's clinical scores in the three evaluation points could not be solely explained by the provision of extrinsic feedback. The finding that five children maintained their scores on the Melbourne Assessment could be explained by the mild severity of impairment of the children in this study. Furthermore, it is not expected to see changes in the Melbourne Assessment after such a short intervention as the amount of practice required to see improvements needs to be larger (Wallen, 2014). On the other hand, the increased scores in the Melbourne Assessment in two children may be explained by the absence of sensory deficits and the high motivation. However, the changes observed in an individual's clinical scores could also be explained by other motor learning principles such as the intensity, the number of repetitions. Furthermore, the occurring development in children might also explain the changes observed in the individual's clinical score. The feedback given was on the remaining time to complete the

task, which could potentially explain the improvement observed in the Jebsen-Taylor Test as this is a timed-test. To be precise, the children received information about how fast they were able to complete the different games proposed during the intervention. Thus, the children focused mostly on trying to complete the game as fast as possible to increase their overall score while trying to be as precise as possible. This may have reflected the observed improvement in the Jebsen-Taylor Test. In other words, without being aware, the children used the speed-accuracy trade-off which is a good paradigm to optimize motor learning (Fitts and Peterson, 1964). This suggests that specific parameters of extrinsic feedback could have an impact on the improvement of upper limb skills. Furthermore, the child with sensory deficits and a MACS level of II was able to improve the time on the Jebsen-Taylor Test. This is an encouraging result since impaired sensation has been previously been linked to reduced motor learning (Robert et al., 2013). This suggests that even with reduced sensation, a child with CP may be able to optimize motor learning when given additional extrinsic feedback.

The variability of the improvement of the kinematic outcomes during the reach-to-grasp task could be explained by several factors. It is important to specify that the range of motion of the shoulder flexion and elbow extension was considered normal in comparison to TD children thus did not need to be improved (Schneiberg et al., 2002). However, this comparison needs to be interpreted with caution as the TD children were younger to those presented in this study. It is clear that the use of VR system as a way to deliver an intervention was able to provide high motivation to the children. However, that alone could not explain why some children were able to improve and others not. Provision of extrinsic feedback could explain some results obtained from this study. For example, feedback given on the excessive use of trunk displacement during the game could explain why most of the children were able to reduce the use of trunk flexion during the reach-to-grasp task. Other factors that could have influenced the variability in kinematic outcomes are motor learning principles such as the intensity, the number of session and the number of repetitions (Kleim and Jones, 2008). As these factors were considered minimal for this research paradigm, especially in comparison to other research paradigms (Gordon et al., 2011), outcome variability was likely. For example, it is often suggested that children with mild impairments require less time of practice in comparison to the children with moderate to more severe impairments to see any changes in the clinical and kinematic outcomes

(Gordon, 2011; Muratori et al., 2013; Novak et al., 2013). However, the minimum effective intensity of practice for children with CP to see a positive effect on motor learning remains unclear.

6.7 Conclusion

In our study, we demonstrated the implementation and acceptability (Bowen et al., 2009) of this research paradigm. This suggests that the use of low-cost, game-based VR system could be used to deliver different frequencies of extrinsic feedback.

Improvement of clinical and kinematic outcomes could be partly attributed to the delivery of extrinsic feedback. Other factors that are known to influence motor learning need to be taken into consideration when it comes to delivering an intervention. Our results suggest that delivering extrinsic feedback through a low-cost, game-based VR system could be used to optimize upper limb motor learning in children with mild hemiplegic CP.

Future studies should investigate other modalities of extrinsic feedback that could optimize motor learning using a game-based VR system. Studies in children with moderate to severe impairments should also be done as the severity of impairments such as reduced sensation and cognitive deficits could negatively influence motor learning (Auld et al., 2012b; McLaughlin et al., 2005; Schmidt and Lee, 2011). Thus, to limit the impact of impairments and reduced sensation, it is suggested to provide additional source of information to optimize motor learning. Particularly, children who have reduced sensation may be able to use other sensory systems to synthesize information from the external world (Dionne-Dostie et al., 2015; Seitz et al., 2006). This concept, defined as multisensory integration, may allow for optimization of motor learning through the integration of multiple sources of information such as intrinsic feedback and additional extrinsic feedback (Dionne-Dostie et al., 2015).

6.8 Limitations

Feasibility in this study only addressed a homogenous group of children with mild impairments, hence results cannot be extended to children with moderate to severe CP with a wider variety of sensorimotor impairments and the presence of cognitive deficits. Interpretation of the results is limited because of the small number of participants. The experimental group was not compared to a control group thus it is difficult to explain why some children were able to improve. Lastly, these results are only applicable to male children with mild hemiplegic CP aged between 11 to 15 years old.

6.9 References

References for this manuscript can be found at the end of the thesis in the Reference section.

Table 6-1 Demographic data and clinical parameters for children with cerebral palsy

Child	Age (y/m)	Side of hemiplegia	MACS	S- W	ROM /24	Proprio /8	Spasticity	MelPre /122	MelPost /122	MelFU /122	JBPre (s)	JBPost (s)	JBFU (s)	IMI /7
C1	11.1	Left	2	2.36	23	8	X1,V3	88	95	105	324.4	290.8	172.6	6.47
C2	12.2	Left	1	2.83	24	8	X1, V2	120	120	120	133.1	91.9	88.8	4.60
C3	14.9	Left	2	2.83	22	8	X2, V2	115	116	117	206.0	222.8	162.6	4.87
C4	12.6	Left	1	2.36	24	8	X1, V3	89	96	119	141.2	182.0	169.6	4.93
F1	13.0	Right	1	2.83	24	8	X1, V3	117	118	116	173.8	104.4	133.5	5.60
F2	13.9	Left	2	3.22	24	8	X1, V3	117	119	119	175.1	139.6	117.7	5.93
F3	13.3	Right	2	4.31	18	8	X2, V2	99	98	100	307.9	241.9	270.4	5.73

Abbreviations : C = Continuous, F = Faded, MACS = Manual Ability Classification System, S-W = Semmes-Weinstein. For S-W, the normal values range is 2.83-3.61 (Bell-Krotoski et al.,1995). ROM = Range of motion., Proprio = Proprioception, X = quality of muscle reaction (maximum score of 5), V = Stretch Velocity, Mel = Melbourne Assessment, FU = Follow-Up, JB = Jebsen-Taylor Test, IMI = Intrinsic Motivation Inventory.

Table 6-2 Improvement in movement time indicated mean differences and effect sizes (ES) for reaches to the close target (2/3 of the arm length), full target (maximal arm length) and far target (maximal arm length plus 2/3).

Table 6-2: Improvement in movement time indicated by mean differences and effect sizes (ES) for reaches to the close target (2/3 of the arm length), full target (maximal arm length) and far target (maximal arm length plus 2/3).

Child no.	Close target				Full target				Far target			
	Postintervention		Follow-up		Postintervention		Follow-up		Postintervention		Follow-up	
	Postintervention baseline difference	ES	Follow-up baseline difference	ES	Postintervention baseline difference	ES	Follow-up baseline difference	ES	Postintervention baseline difference	ES	Follow-up baseline difference	ES
Continuous												
1C	-0.01	-0.07	-0.10	-0.59	0.03	0.22	-0.07	-0.51	-0.03	-0.19	0.00	0.00
2C	-0.02	-0.34	0.07	1.05	0.03	0.30	0.18	1.90	-0.12	-0.64	0.16	0.82
3C	-0.03	-0.32	-0.09	-0.91	0.03	0.41	-0.14	-1.71	0.09	0.48	0.08	0.45
4C	-0.05	-0.43	-0.09	-0.74	-0.10	-0.58	-0.07	-0.39	0.14	0.68	0.08	0.39
Faded												
1F	-0.03	-0.26	0.03	0.35	-0.01	-0.05	0.07	0.55	-0.02	-0.15	0.18	1.14
2F	-0.07	-0.70	0.04	0.45	0.02	0.24	0.03	0.33	0.01	0.07	0.09	1.26
3F	0.02	0.10	-0.10	-0.63	0.16	1.05	-0.04	-0.26	-0.41	-1.35	-0.22	-0.72

Data are shown for two groups of children: continuous or faded feedback. Moderate to large effect sizes indicating improvement are in bold and those indicating deterioration are shaded in grey.

Table 6-3 Improvement in index of curvature indicated by mean differences and effect sizes (ES) for reaches to the close target (2/3 of the arm length), full target (maximal arm length) and far target (maximal arm length plus 2/3).

Table 6-3: Improvement in index of curvature indicated by mean differences and effect sizes (ES) for reaches to the close target (2/3 of the arm length), full target (maximal arm length) and far target (maximal arm length plus 2/3).												
Child no.	Close target				Full target				Far target			
	Postintervention		Follow-up		Postintervention		Follow-up		Postintervention		Follow-up	
	Postintervention baseline difference	ES	Follow-up baseline difference	ES	Postintervention baseline difference	ES	Follow-up baseline difference	ES	Postintervention baseline difference	ES	Follow-up baseline difference	ES
Continuous												
1C	0.21	1.16	-0.09	-	-0.01	-0.08	-0.18	-1.06	0.02	0.13	-0.17	-
2C	-0.07	-0.50	-0.05	-	0.02	0.23	0.05	0.72	-0.01	-0.46	-0.04	-
3C	-0.16	-1.12	-0.20	-	-0.15	-1.85	-0.19	-2.30	-0.08	-0.88	-0.05	-
4C	0.33	4.63	0.15	1.37	0.12	2.97	0.13	3.30	0.04	1.08	0.06	0.56
Faded												
1F	-0.43	-1.08	-0.12	-	-0.21	-0.96	-0.10	-0.46	-0.20	-1.56	-0.08	-
2F	-0.01	-0.13	0.08	0.92	0.07	1.53	0.07	1.50	0.02	0.41	0.05	1.08
3F	-0.04	-0.37	-0.16	-	-0.00	-0.13	-0.11	-2.22	0.03	0.61	-0.10	-
				1.39								2.45

Data are shown for two groups of children: continuous or faded feedback. Moderate to large effect sizes indicating improvement are in bold and those indicating deterioration are shaded in grey.

Table 6-4 Improvement in elbow angle indicated by mean differences and effect sizes (ES) for reaches to the close target (2/3 of the arm length), full target (maximal arm length) and far target (maximal arm length plus 2/3).

Table 6-4: Improvement in **elbow angle** indicated by mean differences and effect sizes (ES) for reaches to the close target (2/3 of the arm length), full target (maximal arm length) and far target (maximal arm length plus 2/3).

Child no.	Close target				Full target				Far target			
	Postintervention		Follow-up		Postintervention		Follow-up		Postintervention		Follow-up	
	Postintervention baseline difference	ES	Follow-up baseline difference	ES	Postintervention baseline difference	ES	Follow-up baseline difference	ES	Postintervention baseline difference	ES	Follow-up baseline difference	ES
Continuous												
1C	14.66	1.81	-20.58	2.54	-16.09	-0.86	-12.45	-0.66	-14.77	-0.72	0.63	0.03
2C	-0.61	-0.07	-6.59	0.74	-11.11	-0.79	-2.02	-0.14	-10.24	-1.27	6.23	0.77
3C	-17.53	-1.33	-14.44	1.09	-20.08	-1.03	-18.98	-0.97	5.68	0.36	4.99	0.31
4C	-9.29	-1.22	-15.69	2.06	5.13	0.26	-4.94	-0.25	27.70	1.44	24.20	1.26
Faded												
1F	3.33	0.22	5.15	0.34	19.01	0.80	24.06	1.01	-10.54	-0.65	-5.92	-0.37
2F	3.08	0.48	6.26	0.97	6.71	1.44	4.64	0.99	-3.34	-0.23	12.12	0.84
3F	5.23	0.65	-3.00	-0.37	-8.94	-0.48	-21.07	-1.13	-12.70	-0.59	-19.67	0.92

Data are shown for two groups of children : continuous or faded feedback. Moderate to large effect sizes indicating improvement are in bold and those indicating deterioration are shaded in grey.

Table 6-5 Improvement in shoulder flexion indicated by mean differences and effect sizes (ES) for reaches to the close target (2/3 of the arm length), full target (maximal arm length) and far target (maximal arm length plus 2/3).

Table 6-5: Improvement in **shoulder flexion** indicated by mean differences and effect sizes (ES) for reaches to the close target (2/3 of the arm length), full target (maximal arm length) and far target (maximal arm length plus 2/3).

Child no.	Close target				Full target				Far target			
	Postintervention		Follow-up		Postintervention		Follow-up		Postintervention		Follow-up	
	Postintervention baseline difference	ES	Follow-up baseline difference	ES	Postintervention baseline difference	ES	Follow-up baseline difference	ES	Postintervention baseline difference	ES	Follow-up baseline difference	ES
Continuous												
1C	-7.11	-1.44	-9.11	1.85	-10.06	-1.15	-7.02	-0.80	-13.19	-0.67	-11.55	0.59
2C	-0.04	-0.01	2.20	0.47	6.53	1.74	7.19	1.91	14.27	3.31	20.55	4.77
3C	-1.60	-0.36	3.81	0.86	6.74	1.78	-4.15	-1.09	7.96	2.24	5.87	1.65
4C	-15.44	-1.15	-7.90	0.59	-17.19	-0.79	-5.48	-0.25	-11.89	-1.03	-14.56	1.26
Faded												
1F	2.02	0.21	11.83	1.25	1.77	0.45	7.40	1.88	-23.40	-0.97	-18.18	0.75
2F	-13.61	-3.18	-4.60	1.07	-13.40	-2.83	-15.04	-3.18	-6.02	-0.89	-2.85	-0.42
3F	-1.60	0.48	-1.71	-0.28	-8.69	-0.99	-20.99	-2.38	-7.65	-0.72	-33.77	3.19

Data are shown for two groups of children: continuous or faded feedback. Moderate to large effect sizes indicating improvement are in bold and those indicating deterioration are shaded in grey.

Table 6-6 Improvement in trunk flexion indicated by mean differences and effect sizes (ES) for reaches to the close target (2/3 of the arm length), full target (maximal arm length) and far target (maximal arm length plus 2/3).

Table 6-6: Improvement in **trunk flexion** indicated by mean differences and effect sizes (ES) for reaches to the close target (2/3 of the arm length), full target (maximal arm length) and far target (maximal arm length plus 2/3).

Child no.	Close target				Full target				Far target			
	Postintervention		Follow-up		Postintervention		Follow-up		Postintervention		Follow-up	
	Postintervention baseline difference	ES	Follow-up baseline difference	ES	Postintervention baseline difference	ES	Follow-up baseline difference	ES	Postintervention baseline difference	ES	Follow-up baseline difference	ES
Continuous												
1C	-2.22	-1.31	3.47	2.06	-5.13	-1.39	7.34	2.00	-7.62	-1.13	17.68	2.61
2C	0.41	1.44	0.92	3.28	-0.49	-0.13	0.58	0.15	-17.38	-1.88	16.02	1.74
3C	-1.14	-1.01	-0.01	-	-1.74	-0.32	-5.40	-0.98	0.74	0.06	7.16	0.62
4C	-0.94	-0.55	-0.04	-	-5.38	-1.23	-9.25	-2.11	-24.71	-3.61	-34.14	-
				0.01								4.98
				0.02								
Faded												
1F	-1.28	-0.29	-1.52	-	9.09	1.98	1.22	0.26	-4.32	-0.50	-4.19	-
				0.49								0.49
2F	0.24	0.07	2.03	0.62	-16.31	-2.69	-9.13	-1.51	-28.10	-2.52	-5.03	-
												0.45
3F	0.76	0.48	2.97	1.87	-0.57	-0.13	3.84	0.86	-8.70	-1.94	17.34	3.86

Data are shown for two groups of children : continuous or faded feedback. Moderate to large effect sizes indicating improvement are in bold and those indicating deterioration are shaded in grey.

Figure 6-1 Research protocol and timeframe of evaluations and training intervention. (A) Timeframe, (B) Name of the games, starting from left to right: Fish Frenzy, Pixel Waves, Catch, Carry and Drop and Pop Clap, (C) Experimental setup for the kinematic assessment.

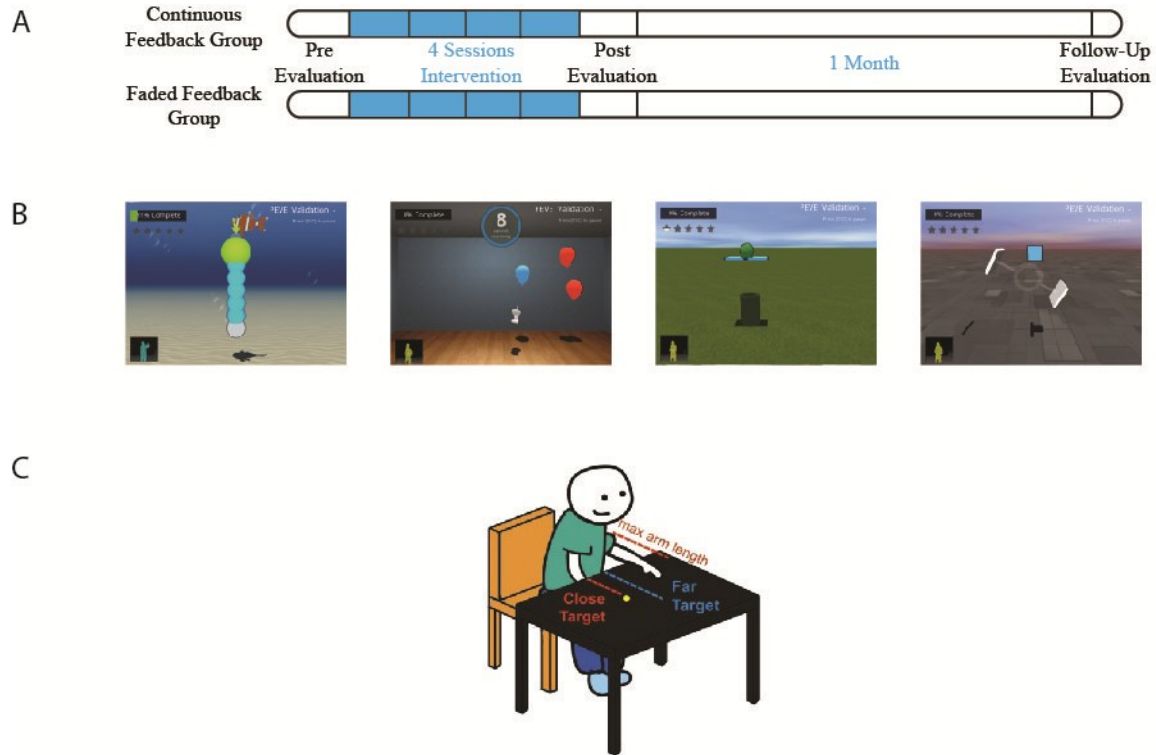


Figure 6-2 Consort flow diagram. Number of participants recruited in this study.

CONSORT 2010 Flow Diagram

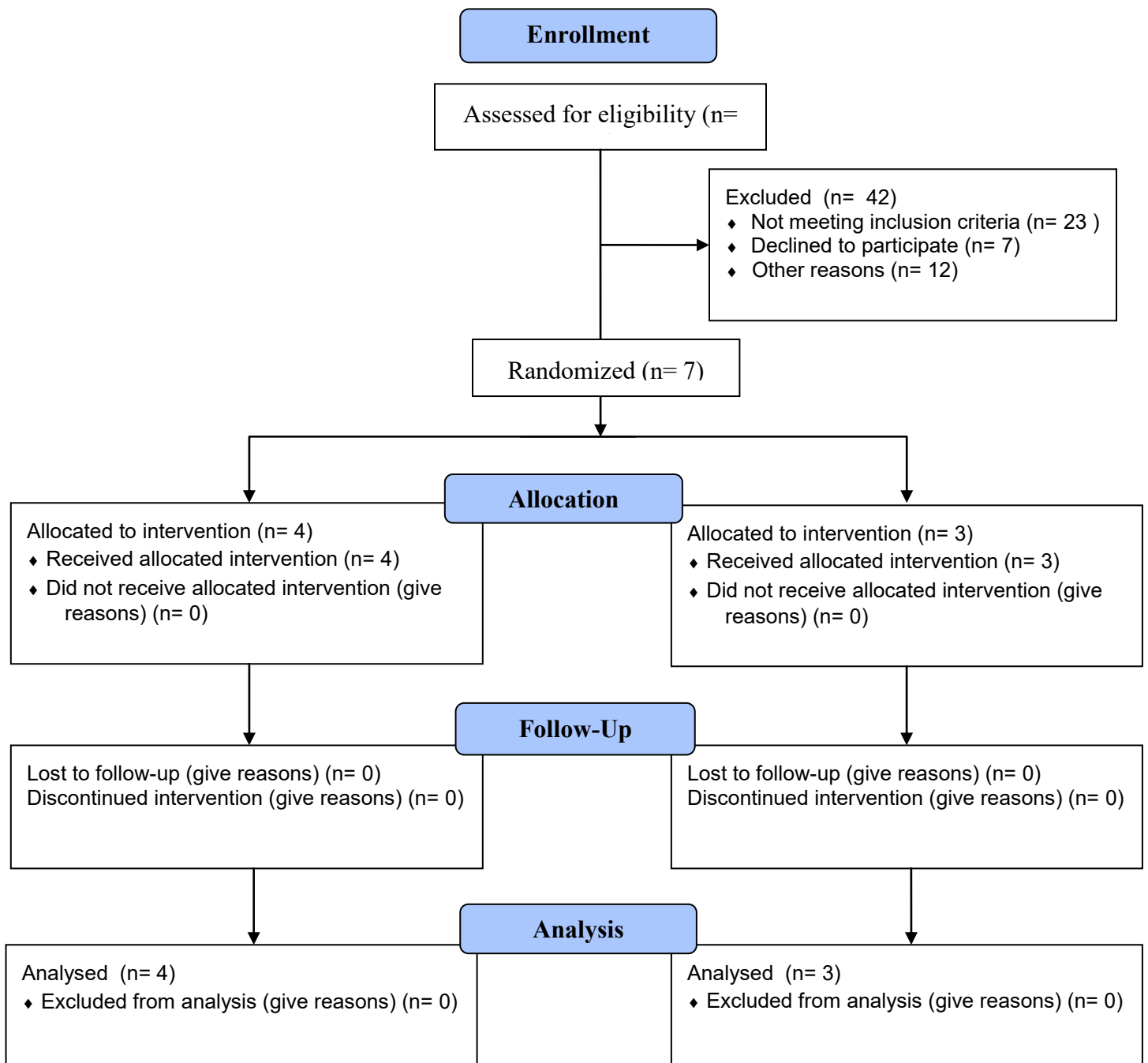
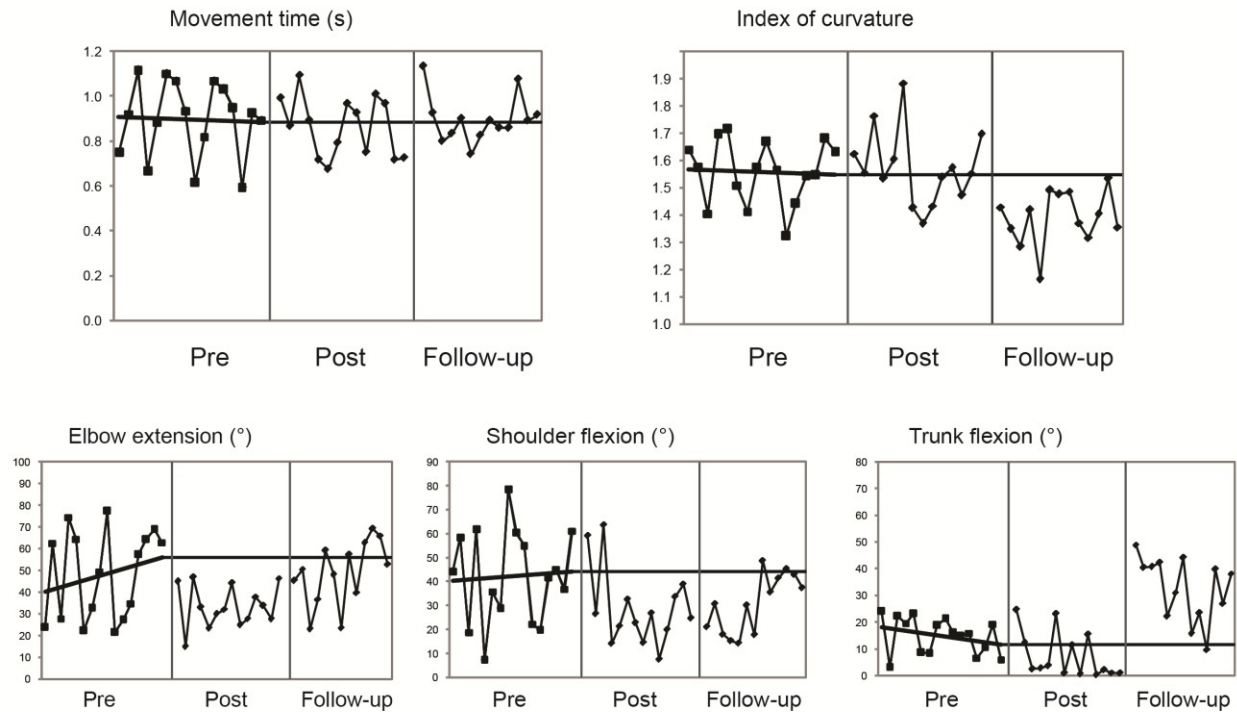


Figure 6-3 Example of trend line analysis. All data points observed in one child for reaches to far target in each phase : baseline (Pre), post-intervention (Post), and follow-up. Data are shown for movement time (far left), trajectory straightness (left), elbow extension (middle), shoulder flexion (right) and trunk flexion (far right).



CHAPTER 7

7.1 General discussion

The global aim of this thesis was to optimize upper limb motor learning through the manipulation of extrinsic feedback in children with hemiplegic CP. The specific objectives were 1) to determine if upper limb kinematics in children with CP during a standardized reach-to-grasp task could be learned, retained and transferred to a similar task, 2) to characterize the role of sensation on motor learning in children with hemiplegic CP, 3) to identify knowledge and gaps on the use of extrinsic feedback to improve upper limb motor skills in TD children and in children with CP, 4) to compare upper limb and trunk kinematics of reaching made in two planes in a 2D virtual environment to those made in a physical environment in TD children and children with CP, 5) to determine the relationships between sensory impairments and reaching kinematics in children with mild hemiplegic CP, 6) to determine the feasibility (i.e., implementation, acceptability) of delivering extrinsic feedback through a VR system for upper limb skill acquisition in children with mild hemiplegic CP, and 7) to determine the feasibility of implementing a short intensive VR intervention in children with mild hemiplegic CP.

These objectives were addressed in four manuscripts featured in this thesis. The first study (Chapter 3) provided evidence that it is feasible to optimize upper limb motor learning through an intervention using a combination of virtual reality training and conventional therapy. This study also showed that children who had better sensation (proprioception and tactile thresholds) learned better in comparison to children who had altered sensation. Kinematic measures during a standardized reach-to-grasp task were used to quantify motor learning. The review (Chapter 4) provided substantial information regarding the importance of providing extrinsic feedback to optimize upper limb motor learning. Results of the third study (Chapter 5) reported the differences in movement performance and quality variables in TD children and in children with mild hemiplegic CP. In children with CP, trajectories were more curved and less trunk flexion and rotation were used in the virtual reality system for all movements. These differences may be related to the user's altered perception and the lack of proper visual cues required to identify object location and distance in the VE. The fourth study (Chapter 6) addressed the feasibility of

an intervention design that delivers different frequencies of extrinsic feedback to improve and to retain upper limb skills following a virtual reality intervention in children with mild hemiplegic CP. Improvements of upper limb motor skills were observed in both clinical and kinematic outcomes.

7.1.1 Importance of Multisensory Integration

Prior the incorporation of the concept of multisensory integration (Section 2.6) in the context of rehabilitation, it is crucial to understand the possible effects of reduced sensation and of any lesion to the somatosensory cortex on motor learning in children with CP. As described in Chapter 2.3.4.4, reduced sensation and lesions in the primary somatosensory cortex were associated with impaired motor learning in animal studies and in healthy adults (Mao et al., 2011; Vidoni et al., 2010). For example, in rodent models, connections between somatosensory cortex (barrel cortex) and the vibrissal motor cortex were identified through different labeling techniques (Mao et al., 2011). These results suggested that the neurons in the superficial layers of the motor cortex couple motor and sensory signals and might mediate sensorimotor integration, which could potentially optimize motor learning.

In healthy adults, Vidoni et al. (2010) investigated the impact of doing repetitive transcranial magnetic stimulation over the primary somatosensory cortex while participants practiced a continuous motor tracking task with the upper limb. Results of the study showed that healthy adults who received repetitive stimulation over the primary somatosensory cortex had reduced wrist proprioception in comparison to the adults who received sham repetitive transcranial magnetic stimulation. In their second experiment, which was the practice of a continuous motor tracking task, both groups improved performance. However, the healthy adults who received repetitive stimulation had less accurate tracking and smaller improvements in performance in comparison to the sham group. These findings suggested that changes associated with learning a new motor skill are limited when the somatosensory cortex is disrupted. This study generally supports the concept that mild to moderate sensory impairment can negatively impact motor learning and that sensory status is an important consideration in motor rehabilitation.

In children with CP, the one study that measured the impact of reduced sensation on motor learning demonstrated that tactile deficits were related to upper limb motor function assessed by the Melbourne Assessment, the Jebsen-Taylor Test and the Assisting Hand Assessment (Auld et al., 2012b). Our results discussed in Chapter 3 were consistent with this finding. After a 15-hour intervention with a pre, post and follow-up evaluation, proprioception and tactile threshold levels were found to be associated with retention of improvements of a standardized reach-to-grasp task.

Thus, to compensate the observed altered sensation in children with CP (Bax et al., 2005; Krigger, 2006), the concept of multisensory integration could be prioritized in order to optimize motor learning. As the multisensory integration concept suggests that the human body is built in a way to facilitate the integration of different sensory inputs (discussed in the next section), the recommendation is to provide additional information through the auditory and visual systems (Lickliter, 2011). This provision of extrinsic feedback may optimize motor learning. However, it is important to emphasize that the provision of extrinsic feedback does not replace the altered sensation; rather, it increases the number of sources of sensory information to promote the optimization of motor learning.

7.1.2 Extrinsic Feedback and Motor Learning

As discussed in Section 7.1.1, the brain facilitates the integration of various sources of information through different sensory systems. Thus, it is suggested that provision of extrinsic feedback will increase the integration of information received through different sensory systems, which may optimize motor learning. The concept of delivering extrinsic feedback was discussed in many theories for its potential to optimize motor learning (Section 2.6.1). For instance, the results of a recent review pertaining healthy adults and individuals with stroke concluded that it was difficult to determine which modality and type of extrinsic feedback are to be prioritized as motor learning depends on a multitude of factors (Sigrist et al., 2013). The complexity of the task, the feedback parameters (KP and KR) and the severity of impairments are therefore all factors to be considered when weighing the cost and the benefit of different feedback modalities.

Results obtained from the third manuscript (Chapter 5) also highlighted the need of a more rigorous and alternative research design to compare the different modalities and frequencies of feedback. Although only a few studies compared auditory and visual feedback, in which all of them reported improvement in learning outcomes, there is no evidence to support any particular modality to be prioritized in both TD children and children with CP. However, visual information is proposed to be more beneficial as it provides more spatial information than auditory information (Witten and Knudsen, 2005). On the other hand, auditory feedback has been recommended for elite athletes as in their circumstances, there is a saturation of visual information (Hermann et al., 2006). This information is to be taken into consideration when it comes to delivering feedback to children with CP as they may rely more on one particular modality, especially in the presence of sensory impairments. Maturation of the different sensory systems also needs to be addressed. During infancy, there is greater reliance on haptic information for motor learning, but as visual and auditory nervous pathways mature, the dependence on visual and auditory sensory modalities increases in order to develop adult-like motor learning strategies (Sigrist et al., 2013; Sullivan et al., 2008). In fact, the auditory and visual cortices are not fully developed until 11 to 13 years of age, which further justifies the reliance on haptic feedback in early motor skill acquisition (Hollants-Gilhuijs et al., 1998; Knoblauch et al., 2001; Kovacs et al., 1999; Moore, 2002).

In healthy adults and in individuals with stroke, faded feedback was suggested to better optimize motor learning in comparison to continuous feedback, although more robust research could confirm this statement (Sigrist et al., 2013). However, these results cannot be extended to children due to their ongoing development (Molier et al., 2010; Sullivan et al., 2008). It is hypothesized that the learner becomes dependent on continuous feedback, upon its availability, in order to improve his/her performance. Thus, the provision of continuous feedback may limit the individual's exploration of different movement possibilities through trial and error and problem solving as seen in the first stage of motor learning in Gentile's theory (Gentile, 1972) (see Section 2.4.1.3 for more information). On the contrary, faded feedback may be more beneficial in comparison to continuous feedback as it allows the exploration of new movement patterns, thus increasing the chance of retaining the motor task due to the involvement of

cognitive processes (Hemayattalab and Rostami, 2010). According to Bernstein's theory of motor learning, there is an abundance of degrees of freedom in early stages of learning. Provision of continuous information in early task learning helps to facilitate the freezing of degrees of freedom, which could potentially have a negative impact on the movement. In other words, receiving this continuous additional information can help the individual to make the decision of which degrees of freedom to focus on while freezing the others. As the second stage of Bernstein's theory of learning gradually releases of degrees of freedom to improve movement skills, the reduction of the feedback frequency should be prioritized in order to incorporate cognitive processes. Faded feedback allows the improvement of the coordination between the different joints or body segments as the individual is not constantly preoccupied with the provided information. Finally, as the individual masters the combination of degrees of freedom, with even less feedback necessary for improvements, the third stage of Bernstein theory of motor learning focuses on the exploitation of the muscle synergies and the use of passive forces. Consistent with these findings and the application of motor learning theories, our results also supported the need to prioritize faded feedback when it comes to a simple task, although more evidence is needed.

For complex tasks, our results also suggest that continuous visual feedback may be more suitable in the first stages of learning as its continuity reduces the cognitive load (Wulf and Shea, 2002). Given that more than two thirds of children with CP have cognitive deficits (Jones et al., 2007), the complexity of the task needs to be considered.

Thus, although suggestions were made with regards to which modalities and frequencies of feedback should be prioritized to optimize upper limb motor learning in children with CP, additional evidence is still needed. Implementation of the research paradigm presented in the fourth manuscript (Chapter 6) was done to compare different frequencies of extrinsic feedback.

7.1.3 Influence of Virtual Environment on Movement Execution

In Section 2.2, movement production was described as being guided by perception of the environment, particularly the object properties, object position and the user's intended action

(Turvey et al., 1977). Perception in a VE may differ from the physical environment as there are limited or altered visual cues. Examples of visual cues include the resolution of the display medium, the use of shadows, drop lines, constancy of the image size. The misinterpretation of those visual cues in a VE may result in altered movements (Kenyon and Afenya, 1995).

There is limited knowledge on how visual cues may alter movements in healthy adults and individuals with stroke as discussed in Section 2.7.4. For example, reaching movements made in a 2D VE by individuals with stroke and in healthy adults were found to be slower, shorter, less straight, less accurate and involved smaller shoulder and elbow joint ranges in comparison to those made in a matched physical environment (Liebermann et al., 2012). Lack of visual cues allowing the interpretation of depth perception may explain this overall decrease in movement quality and movement performance. Another study also found an increase use of elbow extension and decrease use of wrist extension when doing reaching movements in a 2D VE in comparison to those made in a matched physical environment (Viau et al., 2004). Again, these results were attributed to the absence of depth perception and tactile feedback. These differences could also be explained by other factors including the cognitive challenge of interpreting the 3D illusion in the VE, especially when an individual is unfamiliar with the use of virtual reality systems. Moreover, less trunk displacement was observed that is often considered to be a desirable effect since it denotes a reduction in compensatory mechanisms (Cirstea and Levin, 2000; Levin et al., 2009). Reduction of trunk displacement in the VE could be explained by the use of the hand avatar. Indeed, children likely realized that they could successfully reach the virtual targets with an increased range of motion of the shoulder alone while minimizing the use of trunk displacement.

The results obtained in the third manuscript (Chapter 5) tend to go in the same direction as the results presented in the previous paragraph. Indeed, our results indicated that TD children and children with mild hemiplegic CP tended to create more curved trajectories and to require less trunk flexion and rotation for each gesture made in a 2D VE in comparison to those made in a matched physical environment. In contrast to the results obtained in healthy adults and in individuals with stroke (Liebermann et al., 2012), movement times in the frontal and longitudinal plane was not found to be longer in children with CP although differences were observed in TD

children. This absence of difference is possibly explained by the better use of visual cues required for the depth illusion. Furthermore, experience using virtual reality systems may explain the differences observed between children and adults as children are generally more familiar with games.

It is important to be aware of the limitations in order to set appropriate training goals when using a 2D VE for improving upper limb motor learning. Furthermore, the practice of undesired movement may lead to maladaptive plasticity, which is a difficult process to reverse once acquired (Jang, 2013; Nava and Roder, 2011). Maladaptive plasticity occurs when the reorganization of the brain hinders recovery and causes the development of unwanted symptoms (Jang, 2013; Trojan and Pokorny, 1999). An example of maladaptive plasticity was documented in the study by Eyre (2007), which examined neuroplasticity via transcranial magnetic stimulation in infants with either unilateral or bilateral brain lesions. Hypertrophy of the ipsilateral corticospinal tract from the non-infarcted hemisphere was noted, whereas in infants who had bilateral lesions, normal development occurred (i.e., ipsilateral corticospinal tract was pruned by the age of two years). Thus, the authors suggested that both the progressive loss of the contralateral and the strengthening of the bilateral projections of the ipsilateral hemisphere may be considered as maladaptive plasticity. Therefore, although neuroplasticity did occur, an undesirable movement and an increase of functional disability later followed (Eyre, 2007). Moreover, many studies reported that practice that included compensatory movements in individuals with stroke led to poorer outcomes in comparison to those who used what is considered normal movement patterns (Cirstea and Levin, 2000; Krakauer, 2006). In order to minimize the practice of undesired movement, the provision and the manipulation of extrinsic feedback should be used to limit the differences observed in the VE and in the PE. Improvement of the quality of the viewing scene and the use of a better tracking system may also reduce those differences. For example, studies comparing reaching kinematics in a 3D VE to those made in a matched physical environment in healthy adults and in individuals with stroke reported small differences on movement time and trajectory straightness (Knaut et al., 2009; Subramanian et al., 2010). Proper use of visual cues may explain those small differences observed.

7.1.4 Importance of Quantitative Measurements

One of the main issues discussed in our systematic review (see Chapter 4) is the difficulty to interpret the results of Chapter 6 due to the lack of standardized assessment to evaluate improvement of upper limb movements. The majority of the studies reviewed used clinical scales to measure improvement. While clinical scales provide a general idea of motor activities, they lack information about movement quality and movement performance (Subramanian et al., 2010) as well as compensatory mechanisms used to perform the task (Roby-Brami et al., 2003). Clinical scales have also been criticized for their subjectivity, such as in visual assessment during a given task execution (Jaspers et al., 2009; Patterson et al., 2011). Quantitative measurements are generally acceptable to provide advanced information on the movement behavior (Jaspers et al., 2009). In a recent review, kinematic analysis of an upper limb task was proposed to be the gold standard for the evaluation of motion as this assessment provides accurate, reliable, sensitive data to quantify changes in movement quality and movement performance (Visicato et al., 2014). Furthermore, the use of quantitative measurements through kinematics provides a better understanding of compensatory strategies (Visicato et al., 2014). Based on this idea, movement performance and movement quality should be included in the evaluation of treatment effectiveness on motor learning (Levin et al., 2009).

In each study presented in this thesis with the exception of the review, a standardized reach-to-grasp task was used to measure upper limb kinematic changes. This particular task, performed at three distances, is often used in research, because it duplicates the typical behavior of reaching in adults and in children and is associated with the activity capacity level (Alt Murphy et al., 2012). Reaching to a target, which was not practiced during an intervention and is not within the workspace (far target), allows the evaluation of the transferability of the learned task to a similar one. Reaching in a different workspace potentially allows the children to complete other tasks that require a different combination of upper limb ranges of motion.

Kinematic outcomes used in this thesis were restricted to those shown to be reliable to describe arm movements in similarly aged children with CP (Schneiberg et al., 2010). The reliability of six kinematic variables (endpoint trajectory straightness and smoothness, trunk displacement,

elbow extension, shoulder horizontal adduction and shoulder flexion) was tested three times over 5 weeks for each distance. The selected kinematic variables were shown to have moderate to high reliability. Other studies also reported similar results in a similar task in children with hemiplegic CP (Jaspers et al., 2011; Mackey et al., 2005).

Implementation of quantitative measurements helps quantify the effectiveness of the intervention on motor learning and provides additional information on what was learned during the intervention. Lastly, the implementation of a research paradigm in which provision of feedback parameters can be manipulated enables the direct measures of the impact through those kinematic outcomes.

7.1.5 Different Measures to Assess Feasibility

Many clinical studies fail to recruit a sufficient number of participants due to the lack of financial resources and interest of clinicians and families (Bowen et al., 2009). Small studies with low statistical power (i.e., low number of participants recruited) or participants who fail to complete all protocols may lead to a misinterpretation of the data and possibly wasting of valuable resources (Button et al., 2013; Ioannidis, 2005; Schuster and Powers, 2005). Although the average number of participants recruited has not changed over time, the interpretation of the data or findings will likely become more difficult or limited with the increase of study design complexity, which will ultimately require more participants and more assessments (Button et al., 2013). Furthermore, low power leads to difficulty to reproduce and interpret the findings, negatively affecting the validity of findings. Therefore, it is important to consider projects that include a sufficient number of participants as they provide sound results to research questions (Al-Shahi et al., 2014; Button et al., 2013). To assess feasibility, a pilot study is often suggested to determine how likely the proposed research project is to be delivered in the face of all possible limitations and to inform about the readiness of the research design.

According to the National Institute for Health Research (NIH), several key issues should be considered during the development of a feasibility study (Bowen et al., 2009). Their suggestions can be categorized into three themes: clinical relevance, number of participants and logistics at

site. Firstly, the study must be determined to be relevant or not to the clinical community and its surroundings. Secondly, the possibility to recruit a sufficient number of participants is one of the primordial questions in feasibility studies. The availability of a database of the population studied would allow the identification and the collection of additional information for which could possibly explain the obtained results. Examples of supplementary information such as their level of impairments, sensation level, use of feedback and other principles known to have an effect on motor learning in children with CP needs to be considered (Kleim and Jones, 2008). Other considerations include the percentage of eligible participants that is likely to enroll and the expertise of the investigators and, if a pilot study is required. Lastly, the third point raised by the NIH questions the number of sites involved and the availability of the necessary support to conduct such experiments. The researchers also have to keep in mind if the experiments will be beneficial and will accommodate the population studied.

In terms of our feasibility study, acceptability and implementation were two areas of focus in the design of our research paradigm (Chapter 6). The implementation of our research paradigm was considered successful, as all participants were able to complete all the evaluation and the training sessions. Our recruitment rate was considered low to moderate due to problems with the recruitment such as the availability of the participants and their families as well as the small pool of children with CP in this geographic area. However, an implementation of a summer camp or recruitment through the Québec CP Registry would likely increase the number of participants. In regards to the acceptability, all 7 children who participated in this pilot study reported moderate to high motivation during the intervention. High motivation was also reported in previous studies using another VR system in children with CP (Bryanton et al., 2006; Robert et al., 2013). Reasons for high motivation included the fact that the children were engaged in the activity as they felt they were playing video games, rather than undergoing rehabilitation.

7.1.6 Use of Virtual Environment as a Medium to Deliver Feedback to Optimize Motor Learning

The use of VR systems in rehabilitation has increased over the last decades for different populations, particularly in children with CP (Levac et al., 2017). VR systems as an adjunct to

conventional therapy may optimize motor learning because of the increased motivation (Bryanton et al., 2006) and the possibility of manipulating extrinsic feedback (Galvin and Levac, 2011; Riva et al., 2006). Manipulating extrinsic feedback has the advantage of providing precise and consistent information on specific parameters (Schmidt and Lee, 2011). Despite the evidence of the certain advantages of VR systems, there is no general consensus on what is considered the best delivery medium for feedback provision (Molier et al., 2010). This conclusion was also observed in the review included in Chapter 3 and was discussed in-depth in Section 7.1.2.

Before implementing a 2D non-immersive VE (images are projected on the screen) into rehabilitation to optimize motor learning through the manipulation of feedback, it was important to understand if movements are altered by the individual's perception and the visual cues in the VE. The study results in Chapter 5 and in Section 2.7.4 demonstrated that differences were observed in a 2D and a 3D VE when reaching in different planes.

Understanding of the differences present in performance and in movement quality variables facilitates the development of better VR systems, such that in the future, these systems will provide the information on the essential variables. For example, since movements are usually found to be slower in a VE, it is important to provide feedback on the remaining time to complete the task. Furthermore, as observed in the third study (Chapter 5), trajectories tended to be more curved when reaching in the frontal and longitudinal plane, possibly impacting on movement time. Another option that could potentially reduce movement differences is through the manipulation of the variables that are known to alter task difficulty. Such variables include the width of the target and the required distance from point A to point B according to Fitts' Law (Section 2.5.3) (Fitts and Peterson, 1964). For example, it would be possible to reduce the size and to reallocate a target in the VE in order to increase the task difficulty. The manipulation of these variables are also relevant to the Challenge Point Framework, which states that task difficulty should increase with the performance improvement for better motor outcomes (Taub and Wolf, 1997).

Our research paradigm design, which provided extrinsic feedback, considered movement differences observed between a VE and a physical environment. The study presented in Chapter

6 assessed both clinical and kinematic outcomes before, immediately after and one month following a short intensive VR intervention. Participants received concurrent visual and auditory KP and KR feedback after every trial. Participants also received manipulated extrinsic feedback (continuously or faded) at the end of a trial in the form of a combination of visual numerical KP (trunk displacement) and KR (movement precision and velocity). Due to the small sample size, individual data were analyzed.

All participants either maintained or improved their score on the two clinical scales (Jebsen-Taylor Test and Melbourne Assessment). Improvement in both clinical scales could be attributed to the number of repetitions and the intensity of the intervention (Gordon et al., 2008; Kleim and Jones, 2008). However, feedback given on the remaining time could also explain why the participants were able to improve in the Jebsen-Taylor Test, which is considered a timed-test (Jebsen et al., 1969). A standardized reach-to-grasp task (Section 7.1.4) was also used to investigate the effect of feedback on motor learning. Although some children were able to improve either the movement performance or the movement quality, other children were not able to do so. Various explanations could be given as why some children were not able to improve. For instance, since most of the children recruited in this study had mild impairments, there was possibly a ceiling effect. The low number of repetitions and intensity could also explain why some children were not able to improve movement performance. Indeed, some children might need higher intensity and number of repetitions to experience changes.

7.2 Limitations

The limitations of the four studies featured in this thesis were discussed within each respective manuscript. In addition to what was already discussed, for first study, additional measures of sensation would have helped to determine the relationship between sensation and motor learning, such as audition. Furthermore, the correlational analysis between sensation and motor learning may have included too many variables, thus limiting the interpretation of the results. For the second study, the causes of CP could have been discussed in more depth to explain why some children were not able to improve in comparison to others or to TD children. For the third study, the small number of trials analyzed might have negatively influenced the results because of the

kinematic variability when reaching in children with CP. To decrease the reaching variability, more trials should have been recorded and analyzed.. Overall, interpretation of the results of this thesis is limited to children with mild spastic hemiplegic CP. Thus, the results cannot be extended to children who have moderate to severe impairments nor to those who have cognitive deficits. Results of the first, the third and the fourth manuscripts should be interpreted with caution, because the sample sizes were considered low to moderate. Furthermore, the results of the second and the fourth manuscripts can be only applied to 2D non-immersive VE.

None of the studies reported in this thesis have investigated the physiological mechanisms of the use of extrinsic feedback on movement execution and motor learning. Therefore, the results were limited to behavioral changes only.

7.3 Clinical Implications

Most activities of daily living are performed using the upper limbs. In the presence of sensorimotor impairments, reaching may be altered thus limiting the participation level in children with CP. The amount of activities of daily living is directly correlated with quality of life and participation levels in youth and in children with CP (Bjornson et al., 2014). Hence, to increase participation levels, improvement of reaching and activities of the upper limb should be a focus of clinical interventions and can be optimized through the delivery of extrinsic feedback during training.

The current studies demonstrated the importance of measuring sensation in children with CP as sensory deficits may explain why some children are not able to optimize motor learning as efficiently as their healthy peers who do not have reduced sensation. Indeed, improving our understanding of the relationship between reduced sensation and motor learning will help clinicians understand why some children are not able to improve in comparison to their peers without sensory impairments. Furthermore, based on the severity of sensory impairments, clinicians will be able to provide additional information (visual, audition) to compensate for reduced sensation.

Furthermore, results of these studies demonstrated that delivery of additional extrinsic feedback during an intervention is crucial to optimize motor learning but which modalities and types that need to be prioritized remains unknown. However, continuous feedback has been suggested to be efficient, especially when learning a complex task. This additional knowledge and insight on the use of extrinsic feedback in children is important for clinicians as when choosing which frequencies to implement in their intervention.

Clinicians and researchers need to be aware of differences observed in reaching between a physical and virtual environment when setting goals or designing protocols for improving reaching in children with CP using low-cost game-based virtual reality systems. In addition, the last study demonstrated the feasibility of delivering an intervention using a VR system with the possibility of manipulating the delivery of extrinsic feedback and increasing the children's motivation. Consequently, clinicians can use VR systems with the goal of increasing children's motivation and optimizing motor learning through the precise delivery of extrinsic feedback.

7.4 Future directions

Future research is needed to deepen the understanding on the specifics of feedback in general and in the context with VR for children with CP. (1) The efficacy of different frequencies of feedback (i.e., faded vs. continuous) to optimize upper limb motor learning should be further examined. (2) In the goal of improving movement execution and the difference in efficacy between auditory and visual feedback should be characterized. (3) The provision of feedback parameters (i.e., KP and KR) should be studied in relation to measurable improvements of movement quality and/or movement performance. (4) A clinically relevant investigation would need to empirically test the hypothesis whether or not higher intensity and increased repetitions lead to better clinical and kinematic outcomes than the results reported in Chapter 6. (5) The determination of specific optimal combinations of frequencies and types of feedback for a complex task could be clinically pertinent. (6) The efficacy of 3D VR system should be tested for its arm tracking and its use to improve a child's perception of the visual scene, because these two features can ultimately augment the precision of extrinsic feedback.

Future research should address these issues in children with CP who have moderate to severe impairments. Answers to these research questions would help to understand the impact of delivering extrinsic feedback on movement execution and motor learning. The use of neurophysiological assessments may also facilitate the comprehension of the neural-structural changes associated with motor learning when providing extrinsic feedback. Ultimately, the incorporation of the different modalities of extrinsic feedback that optimize motor learning into an intervention may help to improve movement behaviour, which could lead to an increase in functional activity and participation level in children with CP.

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APPENDICES

APPENDIX A. Consent Form for Study 1 of Chapter 3

FORMULAIRE DE CONSENTEMENT À LA PARTICIPATION À L'ÉTUDE INTITULÉE:

L'ÉTUDE « CHILDSTAR » : EFFICACITÉ D'UN ENTRAÎNEMENT À LA STABILITÉ SUR LES FONCTIONS DU BRAS (ATTEINTE ET PRÉHENSION) CHEZ LES ENFANTS AYANT DES DIFFICULTÉS À S'ALIMENTER.

Chercheurs : Heidi Sveistrup, Mindy Levin, Erika Gisel, Patricia McKinley

Étudiante au doctorat : Sheila Schneiberg

Source de fonds : **Les Instituts de recherche en santé du Canada (IRSC)**

Objectifs de la recherche:

L'étude a pour but de déterminer l'efficacité de deux différentes méthodes d'entraînement pour améliorer les fonctions du bras lors des mouvements d'atteinte chez les enfants ayant une infirmité motrice cérébrale (IMC) légère à modérée. L'objectif principal est d'évaluer si un entraînement des tâches d'atteinte et de préhension faites pendant que les mouvements du tronc sont restreints apportera une plus grande amélioration dans l'atteinte et la préhension qu'un entraînement sans restriction physique du tronc.

Nature de la participation de mon enfant

Les enfants seront assignés en vertu d'un tirage pile ou face à l'un de deux groupes d'entraînement. Le groupe 1 recevra un entraînement sur les tâches spécifiques avec instructions verbales du thérapeute concernant l'utilisation du tronc durant l'atteinte. Le groupe 2 recevra un entraînement sur des tâches spécifiques pendant que les déplacements et rotations du tronc seront limités lors des mouvements d'atteinte.

Les participants recevront trois heures d'entraînement par semaine (trois sessions d'une heure par semaine) pour une période de 5 semaines dans un centre de traitement désigné (voir ci-

dessous). De plus, mon enfant participera à 5 sessions de mesure pour évaluer la progression de sa capacité de prendre et manipuler les objets durant l'étude. Les sessions de mesure seront faites 3 fois pendant les cinq semaines précédant l'entraînement, immédiatement après le programme d'entraînement et trois mois après le programme d'entraînement (voir 'Sessions d'évaluations' détaillées ci-dessous).

Ci-dessous, vous trouverez un sommaire du protocole de recherche ainsi que les sites pour les sessions d'évaluation et de l'entraînement :

Période Pré-test			Entraînement	Post-test	Suivi
<i>Semaine 1</i>	<i>Semaine 2.5</i>	<i>Semaine 5</i>	<i>Semaines 6 à 10</i>	<i>Semaine 11</i>	<i>Semaines 24 à 26 (3 mois suivi)</i>
Évaluation 1	Évaluation 2	Évaluation 3	Sessions d'entraînement expérimentales		
¹ Évaluation de laboratoire et fonctionnelle (2 heures)	¹ Évaluation de laboratoire et fonctionnelle (2 heures)	¹ Évaluation de laboratoire et fonctionnelle (2 heures)	3 sessions d'une heure par semaine pendant 5 semaines Groupe 1 : Entraînement sur des tâches avec instructions verbales concernant le mouvement du tronc. Groupe 2 : Entraînement sur des tâches avec restriction physique du mouvement du tronc	<i>Évaluation</i> Post-test ¹ Évaluation de laboratoire et fonctionnelle (2 heures)	Évaluation de suivi ¹ Évaluation de laboratoire et fonctionnelle (2 heures)

¹**Sites pour les sessions d'évaluation et de l'entraînement :**

Institut de réadaptation de Montréal (IRM): Centre de recherche, 4^e étage, 6300 Darlington, Montréal, Qc, Canada, H3S 2J4. Tél : (514) 340-2111 (poste 2188) ou (514) 340-2780.

L'Hôpital juif de la réadaptation : Département pédiatrique; 3205 Place Alton Goldbloom, Laval, Qc, Canada, H7V 1R2. 2^e étage. Tél : (450) 688-9550.

Centre régional de réadaptation La ResSource: 325, rue Laramée, Gatineau (secteur Gatineau), Qc, Canada. Tél : (819) 777-6269.

Sessions d'évaluation

Les sessions d'évaluation seront faites avant l'entraînement, tout de suite après la période d'entraînement et trois mois après la période d'entraînement.

Premièrement, les fonctions du bras de mon enfant seront mesurées par le test « Quality of Upper Extremity Skills » et la spasticité du bras de mon enfant sera aussi mesurée par un test clinique. Ces deux tests prendront environ 30 minutes à compléter.

Deuxièmement, mon enfant fera deux mouvements spécifiques : 1) prendre un morceau de nourriture (morceau de fruit ou de fromage) et l'amener à sa bouche pour le manger; 2) prendre une large morceau de casse-tête et le placer dans un casse-tête devant lui. Durant cette partie de la session, mon enfant sera assis(e) sur une chaise spécialement construite. Ses pieds seront appuyés au sol ou sur un banc et une table sera placée devant lui/elle. La nourriture et le casse-tête seront placés sur la table à différentes distances de mon enfant. Mon enfant fera chacun de ces mouvements vingt-quatre (24) fois et chacun de ses mouvements d'atteinte sera enregistré. Dans le but d'enregistrer le mouvement, des petits marqueurs réfléchissants seront collés sur le bras, la main, la poitrine et le front en utilisant un ruban chirurgical à faible adhérence. Ces séries de tests prendront approximativement 90 minutes à être complétées.

Pendant la session de mesure, des périodes de repos seront planifiées de façon à ce que mon enfant ne devienne pas fatigué(e). Mon enfant pourra se reposer en tout temps à n'importe quel moment à sa demande ou à la mienne.

Sessions d'entraînement

Mon enfant participera dans un programme d'entraînement *orienté vers les tâches* pour les fonctions d'atteinte et saisie de la main. Le programme sera donné par un physiothérapeute ou un ergothérapeute. Le type et la progression des tâches à pratiquer durant les sessions

d'entraînement seront déterminés par le thérapeute et moi/ma famille en concordance avec l'habilité de mon enfant, comme dans le cours normal de la pratique clinique. Les activités peuvent inclure (mais ne sont pas limitées à) l'atteinte, la préhension et la manipulation de jouets de formes variées standards, jouer des instruments de musique, jeux d'ordinateurs ou dessins. Chaque session d'entraînement prendra approximativement une heure.

Engagement des parents

La participation de mon enfant durant cette étude nécessite que j'amène mon enfant au centre _____ pour la prise de mesures, trois (3) fois dans les cinq semaines précédant le programme d'entraînement, une (1) fois immédiatement après la fin du programme d'entraînement, et trois mois après que le programme d'entraînement soit terminé (un total de cinq (5) sessions de mesures). Mon enfant fera la même chose à chacun des jours de tests. Aussi, je dois amener mon enfant au centre _____ trois (3) fois par semaine pendant 15 semaines (15 fois) pour les sessions d'entraînement. Donc, il faut que j'amène mon enfant aux centres pour un total de 20 fois pendant l'étude pour les sessions d'évaluation ou d'entraînement.

Risques et inconvénients découlant de la participation

Malgré le fait que mon enfant ne reçoive aucune thérapie active au moment de l'entrée dans l'étude, un désavantage possible est qu'il (elle) ne pourra pas en recevoir d'autres pendant les 24 à 26 semaines entre les sessions de mesure initiales et finales. Donc, la participation dans l'étude implique que mon enfant ne recevra pas d'autre thérapies pour la durée de l'étude. Cependant, mon enfant peut continuer à effectuer toute activité qu'il/elle fait habituellement à la maison ou à l'école. L'autre désavantage sera le temps qu'il prendra pour effectuer les évaluations durant les périodes de pré-test, de post-test, et de la suivi.

Avantages de participer à cette étude

Il n'y a pas d'avantages directs pour mon enfant quant à sa participation dans cette étude.

Indemnité compensatoire

Les coûts de transport aux centres pour les évaluations pré-test, post-test et suivi seront remboursées jusqu'à un montant maximum de 15,00 \$ par session (total de 5 sessions d'évaluation). Lors des séances du programme d'entraînement, les frais de stationnement seront remboursés (approximativement 5,00 \$ par visite).

Confidentialité de l'information

L'information recueillie dans le cadre des sessions d'évaluations et de l'entraînement sera gardée strictement confidentielle et ne sera utilisée qu'aux fins de la communication scientifique, professionnelle et d'enseignement. Il est entendu que le dossier de mon enfant et toutes autres données concernant mon enfant seront codés de façon à assurer son anonymat. Il est aussi entendu que tous les résultats publiés seront présentés de façon à assurer son anonymat. Tout renseignement personnel (ex. : adresse, numéro de téléphone) sera gardé dans une chemise séparée, accessible seulement par les chercheurs responsables du projet. Cette information pourra être gardée jusqu'à cinq ans après la fin de la période de collecte des données dans un classeur verrouillé au laboratoire de la coordinatrice de l'étude et puis, les données seront détruites à la fin de cette période.

Considérations éthiques

Pour toute question concernant l'éthique de cette recherche ou la participation de mon enfant à cette étude, je peux m'adresser à la coordonnatrice de la recherche au _____, _____ ou _____, la personne en charge des droits des clients.

Questionnement concernant cette étude

Je peux m'enquérir de toutes interrogations concernant cette étude et qu'elles me seront répondues. Ces questions devraient être adressées à Mindy Levin, l'un des chercheurs du projet ou à Sheila Schneiberg, la coordinatrice de l'étude.

Clause de responsabilité

En acceptant de mon enfant participe à cette étude, je ne renonce à aucun de ses droits ni ne libère les chercheurs, leurs commanditaires ou les institutions impliquées de leurs obligations légales et professionnelles.

Personne contact

Pour toute question concernant mes droits ou ma participation à ce projet de recherche, je peux contacter Me Anik Nolet, coordonnatrice à l'éthique de la recherche des établissements du CRIR au 514-527-4527, extension 2643 ou par courriel à l'adresse suivante :
anolet.crir@ssss.gouv.qc.ca

Retrait de l'étude

La participation de mon enfant à l'étude décrite ci-dessus est tout à fait libre et volontaire. Il est également entendu que je pourrai ou que mon enfant pourra, à tout moment, mettre un terme à sa participation sans que cela n'affecte les soins et les services qu'il reçoit ou recevra de l'Hôpital juif de réadaptation ou du Centre de réadaptation La ResSource.

Consentement

Je déclare comprendre ce projet, la nature et le degré de participation de mon enfant et les possibles inconvénients et risques de ce projet tels que décrits dans ce formulaire de consentement. J'ai eu l'opportunité de poser toutes mes questions concernant les différents aspects de cette étude et ai reçu des réponses à ma satisfaction.

Je, _____, soussigné(e), accepte volontairement que mon enfant participe à cette étude. Je peux retirer mon enfant de cette étude en tout temps, sans préjudice. Je certifie que j'ai eu le temps nécessaire pour prendre ma décision et qu'une copie du formulaire de consentement sera placée dans le dossier médical de mon enfant.

Une copie signée de ce formulaire de consentement me sera remise.

Nom du parent

Signature du parent

Date

Nom du témoin

Signature du témoin

Date

Responsabilité du principal chercheur

JE, SOUSSIGNÉ(E), _____, CERTIFIE QUE :

(A) J'AI EXPLIQUÉ AU PARTICIPANT LES TERMES DE CE PRÉSENT FORMULAIRE DE CONSENTEMENT;

(B) J'AI RÉPONDU À TOUTES LES QUESTIONS QUI M'ONT ÉTÉ POSÉES ;

(C) J'AI CLAIREMENT INDIQUÉ AU PARENT DE L'ENFANT QU'IL PEUT RETIRER SON ENFANT DE L'ÉTUDE EN TOUT TEMPS; ET (D) QUE JE LUI DONNERAI UNE COPIE SIGNÉE ET DATÉE DE CE FORMULAIRE.

Signature du coordonnateur

Date

Mindy Levin

Université McGill

École de physiothérapie et d'ergothérapie (514) 398-3994

Hôpital juif de réadaptation (450) 688-9550, poste 3834

Institut de réadaptation de Montréal (514) 340-2780

Sheila Schneiberg

Université McGill

École de physiothérapie et ergothérapie

Hôpital juif de réadaptation (450) 688-9550, poste 3834

Institut de réhabilitation de Montréal (514) 340-2111, poste 2188

Patricia McKinley

Université McGill

École de physiothérapie et d'ergothérapie (514) 398-4498

Centre de réadaptation Constance-Lethbridge (514) 487-1891, poste 352

Erika Gisell

Université McGill

École de physiothérapie et d'ergothérapie (514) 398-4510

Heidi Sveistrup

Université d'Ottawa

Faculté des Sciences de la Santé (613) 562-5800

APPENDIX B. Consent Form for Study 3 of Chapter 5

CONSENT FORM FOR PARTICIPATING IN A RESEARCH PROJECT

STUDY TITLE

Validation of arm movements in virtual reality in children and youth with Cerebral Palsy.

PRINCIPAL INVESTIGATORS

Mindy F. Levin, PT, Ph.D., McGill University

Maxime Robert, M.Sc., Ph.D. Candidate, McGill University

INTRODUCTION

We are asking your child to participate in a research project to help us determine how reaching movements made while playing a computer game in a 2D virtual reality environment may differ from those made in a physical environment. Before agreeing to participate in this project, please take the time to read and carefully consider the following information.

This consent form explains the aim of this study, the procedures, advantages, risks and inconveniences as well as the persons to contact if you have any questions or concerns.

This consent form may contain words that you do not understand. We invite you to ask the research team to explain anything that is not clear to you.

DESCRIPTION OF THE STUDY AND ITS PURPOSE

The use of virtual reality (VR) applications (e.g., games) to encourage people to move more has been increasing over the last decade in rehabilitation, especially in children. We have shown previously that the movements executed by adults in different types of virtual environments can be similar to those made in physical environments although some differences were observed. To date however, we do not know whether movements made by children playing VR games, differ from movements made in a natural environment. This project will evaluate the similarities and

differences in reaching movements made by children with Cerebral Palsy in these two environments.

Special equipment to do exercise and to record arm movements will be used in this study. They are a VR game-like exercise system, a physical environment and an electromagnetic system to measure arm movement. Those three systems have already been used in many studies in our laboratory. The VR system will encourage different reaching movements in several game-like scenarios. These movements will be repeated in a physical environment. We will record your child's arm movements while interacting with each of these environments.

NATURE AND DURATION OF YOUR PARTICIPATION

Your child is invited to participate in the research project which entails only one evaluation session lasting approximately one hour. The session will take place at the Jewish Rehabilitation Hospital and includes presentation of the study and evaluation time.

Evaluation session

If your child accepts to participate in this study, you will be asked to respond to brief questions about your child's medical history. This information will be used to determine if there are any medical or scientific reasons why your child should not participate in this study.

a) Clinical Evaluation: Your child will be asked to answer a questionnaire to evaluate his/her handedness using an established clinical scale. We will also measure how your child uses his/her arm with an established clinical functional scale. For this test, your child will be asked to perform a series of actions to test arm function. These tests will be performed by a clinician

b) Reaching Evaluation: How your child coordinates the movements of his/her arms and trunk when reaching from a sitting position will be measured. Your child's movements will be recorded with an electromagnetic system by 6 markers attached with hypoallergenic tape to the skin of the arms and trunk. Your child will reach with his/her arm to targets placed in front of him/her at 3 different distances. He/she will repeat each movement 15 times for a total of 45

movements. He/she will repeat the same 45 movements in the other environment for a total of 90 movements.

PERSONAL BENEFITS FROM PARTICIPATING IN THE RESEARCH STUDY

Your child will personally not benefit from taking part in this study. However, your child might helps us by making a contribution to the advancement of science in the field of understanding the movements performed in both virtual and physical environments.

RISKS ASSOCIATED WITH PARTICIPATING IN THE RESEARCH STUDY:

Your child's participation in this research study involves no risk. It is understood that your child's participation in the study will not affect the care and services, which your child currently receives or will receive.

INCONVENIENCES ASSOCIATED WITH PARTICIPATING IN THE RESEARCH STUDY:

It is possible that the effort exerted during the evaluation may cause fatigue but this will be temporary. It is understood that if your child becomes tired during the evaluation, he/she will be able to rest at any time before continuing.

The travel time from your home to the research centre and the evaluation time may represent an inconvenience for some children or their families.

ACCESS TO YOUR CHILD'S MEDICAL CHART

You grant access to your child's medical records to the investigators of this project for the purpose of collecting pertinent information (lesion type, medical history, medications, upper limb motor abilities, perceptual and/or cognitive ability).

CONFIDENTIALITY

All personal information gathered about your child during the study will be coded in order to ensure your child's confidentiality. Only the members of the research team will have access to this information. However, in order to monitor the research project, the research records of your child may be consulted by a person mandated by the Research Ethics Committee of CRIR or by

an Ethics Unit of the Minister of Health and Social Science of Quebec, who adhere to a policy of strict confidentiality. This data will be kept under lock and key at the Jewish Rehabilitation Hospital by the person in charge of this study for a period of five years following the end of the study, after which it will be destroyed. In the event that the results of this study are presented or published, no information identifying your child will be included.

QUESTIONS CONCERNING THE STUDY

For your satisfaction, we will take care of answering all the questions you have regarding this project to which your child is agreeing to participate. For information or questions, please contact the persons in charge of this project: Maxime Robert at 450-688-9550 extension 4824 or Mindy Levin at 450-688-9550 extension 3834.

VOLUNTARY PARTICIPATION AND WITHDRAWAL OF YOUR CHILD'S PARTICIPATION

Participation of your child in this research study is completely voluntary. It is understood that your child or yourself can, at any time, withdraw his/her participation without this affecting the health care and services he/she is receiving or will receive from institutions in CRIR.

In case of withdrawal from the study, all documents concerning your child will be destroyed if that is your decision.

RESPONSIBILITY CLAUSE

By agreeing to participate in this study, you do not give up any of your child's legal rights nor release the researchers, sponsors or institutions involved of their legal and professional obligations.

COMPENSATORY INDEMNITY

A compensatory indemnity will be provided to compensate you for any inconveniences or expenses associated with your child's participation (traveling and parking) up to a maximum of \$10 for the evaluation after presentation of receipts (if applicable).

CONTACT PERSON

If you have any questions about your rights and recourse or your participation in this research study, you can contact Me Anik Nolet, Research Ethics Co-ordinator for the CRIR'S Institutions at (514) 527-4527 extension 2649 or by e-mail: anolet.crir@ssss.gouv.qc.ca. For these questions, you can also contact the Ombudsman person of the Jewish rehabilitation Hospital.

CONSENT

I state that I have read this consent form. I understand this study, the nature and extent of my child's participation, as well as the benefits and risks/inconveniences to which my child will be exposed as presented in this form. I have been given the opportunity to ask questions concerning any aspects of the study and have received answers to my satisfaction.

I, the undersigned, voluntarily agree that my child may take part in this study. My child can withdraw from the study at any time without prejudice of any kind. I certify that I have had sufficient time to consider my decision that my child may participate in this study. A copy of this consent form will be placed into my child's medical chart.

A signed copy of this consent form will be given to me.

I accept to be contacted by the same researchers for my child to participate in other scientific studies done in a similar area of research.

Yes: a) for one year ☐ b) for two years ☐ c) for three years ☐ No ☐

NAME OF PARTICIPANT (print)

NAME OF LEGAL REPRESENTATIVE COMPULSORY SIGNATURE OF LEGAL
OF MINOR PARTICIPANT LEGAL REPRESENTATIVE
(print)

Signed at _____, the _____, 201____.

COMMITMENT OF THE RESEARCHER

I, the undersigned, _____, certify
(print)

- (a) having explained to the research participant the terms of this form
- (b) having answered all the questions he/she as asked in this regard
- (c) having clearly indicated that he/she remains free, at any time, to end his/her participation in the above described research study
- (d) that I will give him/her a signed and dated copy of this form

Signature of the Principal Investigator
or representative

Signed at _____, the _____ 201__.

APPENDIX C. Consent Form for Study 4 of Chapter 6

Formulaire de consentement pour les participants d'un projet de recherche

TITRE DU PROJET :

Le rôle du feedback dans l'apprentissage moteur chez les enfants avec une paralysie cérébrale.

RESPONSABLES DU PROJET

Mindy F. Levin, pht, Ph.D., Université McGill

Maxime Robert, M.Sc. Candidat au doctorat, Université McGill

Krithika Sambasivan, B.Sc, M.Sc., Candidate au doctorat, Université McGill

PRÉAMBULE

Nous sollicitons la participation de votre enfant à un projet de recherche dans le but de déterminer la quantité de feedback à donner aux enfants afin d'améliorer les mouvements des bras. Le feedback donné est une rétroaction sur la performance du participant. Avant d'accepter de participer à ce projet de recherche, prenez le temps de comprendre et de considérer attentivement les renseignements qui suivent.

Ce formulaire de consentement vous explique le but de cette étude, les procédures, les avantages, les risques et inconvénients, de même que les personnes avec qui communiquer si vous avez des questions ou des inquiétudes.

Le présent formulaire de consentement peut contenir des mots que vous ne comprenez pas. Nous vous invitons à poser toutes les questions pour clarifier ce qui n'est pas clair pour vous.

DESCRIPTION DU PROJET ET DE SES OBJECTIFS

Les enfants avec la paralysie cérébrale peuvent éprouver de la difficulté à bouger leurs bras. Dans les activités de la vie quotidienne, ces incapacités font qu'ils éprouvent plusieurs difficultés notamment pour saisir un objet comme prendre un crayon pour ne donner qu'un seul exemple. Nous savons que les mouvements d'atteintes peuvent être améliorés chez les enfants à

développement typique lorsqu'un nombre spécifique de feedback est donné durant un exercice. Ce projet va évaluer quel est le meilleur feedback pour améliorer les mouvements de bras chez les enfants avec la paralysie cérébrale.

Dans cette étude, des équipements spéciaux pour faire les exercices et pour enregistrer les mouvements seront utilisés. Ces équipements sont un système de réalité virtuel pour les exercices et un système électromagnétique pour enregistrer les mouvements des bras. Ces deux systèmes ont été utilisés à maintes reprises dans plusieurs études de notre laboratoire. Le système de réalité virtuel nous permettra de modifier la fréquence et le type de feedback donné durant les exercices. Après une évaluation sur la manière que votre enfant effectue des mouvements avec ces bras, il sera placé au hasard dans un des trois groupes en jouant à des jeux vidéo sur un système de réalité virtuel impliquant la pratique des mouvements. La participation de votre enfant inclut 3 sessions d'évaluation et 4 sessions d'exercices étalés sur une période de 7 jours.

NATURE ET DURÉE DE LA PARTICIPATION :

Votre enfant est invité à participer à ce projet de recherche. Pour les évaluations, une évaluation aura lieu avant, une après les exercices et la troisième évaluation aura lieu un mois plus tard. Il y aura 4 sessions d'exercice. Chaque session a une durée approximative de 2 à 3 heures et aura lieu soit à l'Hôpital Juif de Réadaptation, au Centre de Réadaptation Marie Enfant ou à votre domicile si c'est possible.

La participation de votre enfant implique :

Première visite : Pré-test

Cette session a une durée approximative d'environ 3 heures, incluant le temps de présentation de l'étude et le temps d'évaluation.

Si votre enfant accepte de participer, nous vous demanderons de répondre à des questions au sujet de ses antécédents médicaux. Ces documents serviront entre autres à déterminer s'il existe des causes médicales ou scientifiques pour l'exclusion de votre enfant à cette étude.

I.

a) Évaluation fonctionnelle : Nous mesurerons la longueur du bras de votre enfant, sa sensation, sa spasticité, sa latéralité et sa motivation en utilisant des échelles cliniques établies. Nous allons aussi mesurer comment votre enfant utilise ses bras avec des échelles cliniques fonctionnelles. Pour ce test, enregistré sur vidéo, nous demanderons à votre enfant d'effectuer une série de mouvements afin d'évaluer les fonctions de son bras. Ces tests seront effectués par un clinicien.

b) Évaluation de tâche d'atteinte: En position assise, la coordination des mouvements des bras et du tronc de votre enfant sera enregistrée. Six réflecteurs seront apposés à l'aide d'un autocollant hypoallergène sur la peau du bras et du tronc de votre enfant afin d'enregistrer ses mouvements. Votre enfant devra effectuer trois conditions distinctes afin d'atteindre avec son bras une cible située devant lui. Votre enfant devra répéter ces mouvements 15 fois pour chaque mouvement pour un total de 45 mouvements.

II. De la 2e session à la 5e session – sessions d'exercice

Les sessions d'exercice auront une durée approximative de deux heures chacune pour un total de 8 heures.

Intervention : On demandera à votre enfant de jouer à quatre jeux différents en utilisant un système de réalité virtuel. Les deux premiers jeux consistent à suivre une trajectoire avec les mouvements du bras ou de toucher un ballon afin de compléter les jeux. Pour les deux autres jeux, la jouabilité consiste à attraper des objets avec les deux mains. Une caméra (Kinect, Microsoft) captera les mouvements des bras de votre enfant. Chaque session d'exercice aura une durée de 2 heures. Une période de repos sera donnée après chaque jeu et après 30 minutes de jeu. Votre enfant sera placé au hasard dans un des trois groupes auquel la fréquence de feedback va varier. Dans le premier groupe, aucun feedback additionnel ne sera donné à votre enfant. Dans le deuxième groupe et le troisième groupe, en plus de recevoir du feedback visuel lors des jeux, un nombre spécifique de feedback additionnel sera donné. Par exemple, dans le deuxième groupe, un feedback additionnel sera donné après chacun des mouvements. Pour le troisième groupe, le feedback additionnel sera donné après un certain nombre de mouvements.

Après la première session d'exercice, nous allons répéter la section « Évaluation de tâche d'atteinte (Ib)» tel que décrite ci-dessus.

Sixième visite : Post-test

Nous allons répéter les évaluations Ia et Ib.

Septième visite : Follow-up (Un mois)

Nous allons répéter les évaluations Ia et Ib.

AVANTAGES POUVANT DÉCOULER DE VOTRE PARTICIPATION :

Votre enfant ne retire personnellement aucun avantage à participer à cette étude. Dans certain cas, la mobilité du membre supérieur de votre enfant pourrait s'améliorer. En plus de contribuer à l'avancement de la science l'information recueillie de votre enfant lors de cette étude nous aidera à comprendre les mécanismes d'optimisation de l'apprentissage moteur chez les enfants avec la paralysie cérébrale.

RISQUES POUVANT DÉCOULER DE VOTRE PARTICIPATION :

La participation de votre enfant à ce projet de recherche ne comporte aucun risque que ce soit. Il est aussi entendu que sa participation n'affectera pas les soins et les services qu'il reçoit ou recevra.

INCONVÉNIENTS PERSONNELS

Il se peut que les efforts demandés lors de l'évaluation provoquent une certaine fatigue mais celle-ci ne sera que temporaire. Il est entendu que si votre enfant est fatigué durant n'importe quelle session, il pourra se reposer en tout temps avant de continuer.

Le déplacement de votre domicile au site de recherche et la durée des évaluations/exercices peuvent représenter pour certains enfants ou familles un inconvénient.

ACCÈS AU DOSSIER MÉDICAL DE VOTRE ENFANT

Vous acceptez que les personnes responsables de ce projet aient accès au dossier médical de votre enfant afin de recueillir des informations pertinentes (type de lésion, antécédent médicaux, médications, habilité motrice des membres supérieurs, perception et/ou habilité cognitive).

CONFIDENTIALITÉ :

Tous les renseignements personnels recueillis au sujet de votre sujet au cours de l'étude seront codifiés afin d'assurer leur confidentialité. Seuls les membres de l'équipe de recherche y auront accès. Cependant, à des fins de contrôle du projet de recherche, le dossier de recherche de votre enfant pourrait être consulté par une personne mandatée par le CÉR des établissements du CRIR ou par l'Unité de l'éthique du ministère de la Santé et des Services sociaux du Québec, qui adhère à une politique de stricte confidentialité. Ces données ainsi que les enregistrements seront conservées sous clé à l'Hôpital juif de réadaptation par le responsable de l'étude pour une période de 5 ans suivant la fin du projet. Après quoi, elles seront détruites. En cas de présentation de résultats de cette recherche ou de publication, rien ne pourra permettre d'identifier votre enfant.

QUESTIONS CONCERNANT CETTE ÉTUDE

Pour votre satisfaction, nous nous appliquerons à répondre à toutes questions que vous poserez à propos du projet de recherche auquel vous acceptez de participer. Pour toutes informations ou questions, vous pourrez communiquer avec les personnes suivantes : Maxime Robert au (450) 688-9550 poste 4824 ou Mindy Levin au (450) 688-9550 poste 3834.

PARTICIPATION VOLONTAIRE ET RETRAIT DE LA PARTICIPATION DE VOTRE ENFANT

La participation de votre enfant au projet de recherche décrit ci-dessus est tout à fait libre et volontaire. Il est entendu que votre enfant ou vous-mêmes pourront, à tout moment, mettre un terme à sa participation sans que cela n'affecte les soins et les services de santé que votre enfant reçoit ou recevra de l'hôpital juif de réadaptation.

En cas de retrait de la part de votre enfant, tous les documents audiovisuels et écrits le concernant seront détruits si vous le souhaitez.

CLAUSE DE RESPONSABILITÉ :

En acceptant de participer à cette étude, votre enfant ne renonce à aucun de ses droits ni ne libère les chercheurs, le commanditaire ou les institutions impliquées de leurs obligations légales et professionnelles

INDEMNITÉ COMPENSATOIRE :

Une indemnité compensatoire vous sera remise afin de défrayer les coûts et les inconvénients associés à la participation de votre enfant (frais de transport et stationnement) jusqu'à un maximum de \$30 par séance d'évaluation pour un maximum de 210\$, après d'avoir présenté les reçus (si applicable).

PERSONNES-RESSOURCES

Si vous avez des questions sur vos droits et recours ou sur votre participation à ce projet de recherche, vous pouvez communiquer avec Me Anik Nolet, coordonnatrice à l'éthique de la recherche des établissements du CRIR au (514) 527-4527 poste 2649 ou par courriel à l'adresse suivante: anolet.crir@ssss.gouv.qc.ca. Pour ces questions, vous pouvez également communiquer avec le commissaire local aux plaintes de l'Hôpital juif de réadaptation.

CONSENTEMENT

Je déclare avoir lu et compris le présent projet, la nature et l'ampleur de ma participation, ainsi que les risques auxquels mon enfant s'expose tels que présentés dans le présent formulaire. J'ai eu l'occasion de poser toutes les questions concernant les différents aspects de l'étude et de recevoir des réponses à ma satisfaction.

Je, soussigné(e), accepte volontairement que mon enfant participe à cette étude. Mon enfant peut se retirer en tout temps sans préjudice d'aucune sorte. Je certifie qu'on m'a laissé le temps voulu pour prendre ma décision pour que mon enfant participe à ce projet.

Une copie signée de ce formulaire d'information et de consentement doit m'être remise.

J'accepte d'être contacté(e) par la même chercheure pour participer à d'autres études scientifiques menées dans un domaine de recherche similaire:

Oui a) pour un an ☐ b) pour deux ans ☐ c) pour trois ans ☐

Non ☐

NOM DU PARTICIPANT _____

NOM DU REPRÉSENTANT _____

SIGNATURE OBLIGATOIRE LÉGAL DU PARTICIPANT DU REPRÉSENTANT LÉGAL
MINEUR _____

Fait à _____ le _____, 20____.

ENGAGEMENT DU CHERCHEUR

Je, soussigné(e), _____, certifie

(a) avoir expliqué au signataire les termes du présent formulaire;

(b) avoir répondu aux questions qu'il m'a posées à cet égard;

(c) lui avoir clairement indiqué qu'il reste, à tout moment, de mettre un terme à sa participation au projet de recherche décrit ci-dessus; et

(d) que je lui remettrai une copie signée et datée du présent formulaire.

Signature du responsable du projet

ou de son représentant

Fait à _____, le _____ 20____.