Rehabilitation strategies to improve upper limb movement quality in children with cerebral palsy

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A thesis submitted to the Faculty of Graduate Studies and Research in partial fulfillment of the requirements of the degree of Doctorate in Philosophy (Rehabilitation Science)

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DEDICATION

I dedicate this doctoral thesis to the memory of my beloved niece, the late Isabella Caroline Coriolano Valença Dias - ‘Belinha’. The memory of your smile, courage and strong, yet innocent faith in our Lord Jesus, inspired me to strengthen my own faith. Because of your example, I was reassured, knowing that I was guided and protected throughout this journey towards obtaining my doctorate from beginning to end.

My faith, the memory of Belinha, and my passion for pediatric rehabilitation served as my true sources of strength, perseverance and motivation to complete this thesis.
“Everything we do, it is a drop in the ocean, but if we do not do it, it will be missing forever” - Mother Theresa of Calcutta
ACKNOWLEDGMENTS

The accomplishment of a PhD thesis cannot be credited to an individual alone; its achievement involves many people. Completing a thesis can be likened to a long-distance run – like that of Canadian icon and hero Terry Fox – where sheer perseverance and endurance play a major role. The winner is not necessarily the one who arrives first, but the one who completes the competition. In a long intellectual journey each obstacle surpassed represents a new threshold of learning. An endeavour such as this is not possible without the aid of special coaches. I was fortunate enough to have two experts to guide me: my supervisor Dr. Mindy Levin and my co-supervisor Dr. Patricia McKinley.

Dr. Levin is the first person to whom I want to give thanks because it was her who introduced me to scientific research. I owe to her everything that I have learned about conducting experimental research in rehabilitation. Her expertise in physiology, motor control and the way that her research links those domains with rehabilitation reaffirm that I made the right choice coming to Canada, and choosing her as my supervisor. Dr. Levin’s attention to minute details and her persistence in achieving perfection has taught me how to apply a critical appraisal of scientific literature using her standards of research. All who get the chance to know Dr. Levin are impressed by her multitasking abilities and pragmatic approach to scientific research. Through her I learned that science is not just about having ideas or good marks, but meeting deadlines and complying with department policies as well. Mindy, I cannot put into words the gratitude I feel for you for all the support that you have given me throughout these years. Thank you.

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sabbatical, Dr. McKinley still found time to devote herself to helping me achieve all steps of this thesis and its accompanying papers. Pat, I thank you from the bottom of my heart.

In a long run, sometimes the effort put forth is so intense that you cannot give anymore. Suddenly though, you look ahead and see a gentle and familiar face at the end of the race line, giving you that last boost of energy that you need to complete the race. In my case, this gentle and familiar face was Dr. Nancy Mayo. Dr. Mayo was an enormous support, especially in the last part of this thesis. She generously imparted to me her epidemiology expertise and encouraged me to apply a single subject design to this project. However, most importantly, Dr. Mayo, through her kindness and passion for rehabilitation, reminded me that there are many resources to prove treatment efficacy, and by showing me that, she reaffirmed my own passion in rehabilitation research. Dr. Mayo, thank you so very much for your encouragement and most especially for helping me get back on track when I began to stray or lose heart.

Even a marathon runner has to be reminded of how close she is to the finish line. Dr. Susan Bartlett kept reminding me that I had to finish. I did not understand her persistence on this point, but only now do I realize the worth of it. Dr. Bartlett, I thank you deeply from my heart.

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I would not have been able to conclude this thesis without the support and company of my friends. They are: Sibele Melo, Helli Raptis, Youssef Bououlid Idrissi, Fabiana Antunes, Eros Oliveira, Stella Maris Michaelsen, Ksenia Ustinova, Luis Fernando Requiao, Luiz Alberto Knault, Rubens Silva, Saandeep Subraamanian, Archana Sangole, Nadine Kaseka Musampa. My ever patient room-mate, who accommodated my expanding occupation of our shared living room and who was a source of peace and friendship during challenging times with this thesis: Melanie Banina, I thank you. I would like to thank my neighbor Angely Pacis for providing me with moral support and laughter. Lastly, I want to thank Anne Marie Hong Van Le who has fed me with her special *Tonkinois* soup throughout this writing process and who has warmed my soul with her good thoughts and words.

Finally, I owe everything that I am, and everything that I have achieved to God, the Blessed Virgin Mary and my parents, brothers, nieces and nephews. My parents taught me that with hard work everything is possible. José Arlindo Valença Dias and Eliete Schneiberg Valença Dias, muito obrigada – Eu amo vocês.
STATEMENT OF AUTHORSHIP

I certify that I am the primary author of all manuscripts contained in this thesis. I claim full responsibility for the content and style of the text here included.
<table>
<thead>
<tr>
<th>Abbreviation</th>
<th>Description</th>
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<tbody>
<tr>
<td>CNS</td>
<td>Central Nervous System</td>
</tr>
<tr>
<td>CP</td>
<td>Cerebral Palsy</td>
</tr>
<tr>
<td>CSI</td>
<td>Composite Spasticity Index</td>
</tr>
<tr>
<td>EBM</td>
<td>Evidence-Based Medicine</td>
</tr>
<tr>
<td>EBP</td>
<td>Evidence-Based Practice</td>
</tr>
<tr>
<td>FR</td>
<td>Frame of Reference</td>
</tr>
<tr>
<td>GMFCS</td>
<td>Gross Motor Function Classification System</td>
</tr>
<tr>
<td>IC</td>
<td>Index of Curvature</td>
</tr>
<tr>
<td>ICF</td>
<td>International Classification of Functioning, Disability and Health</td>
</tr>
<tr>
<td>IRED</td>
<td>Infra-Red Emitting Diodes</td>
</tr>
<tr>
<td>MACS</td>
<td>Manual Ability Classification System</td>
</tr>
<tr>
<td>mCIT</td>
<td>Modified Constraint Induced Therapy</td>
</tr>
<tr>
<td>NTR</td>
<td>No Trunk Restraint</td>
</tr>
<tr>
<td>PEDI</td>
<td>Pediatric Evaluation of Disability Inventory</td>
</tr>
<tr>
<td>PROM</td>
<td>Passive Range of Motion</td>
</tr>
<tr>
<td>SSRD</td>
<td>Single Subject Research Design</td>
</tr>
<tr>
<td>TD</td>
<td>Typically Developed</td>
</tr>
<tr>
<td>TR</td>
<td>Trunk Restraint</td>
</tr>
<tr>
<td>WeeFIM</td>
<td>Functional Independence Measure</td>
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<td>WHO</td>
<td>World Health Organization</td>
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ABSTRACT

Children with CP are extremely heterogeneous in terms of etiology and clinical features. The diversity of symptoms among CP syndromes is a challenge for different branches of health research. Despite the efforts of many studies in examining rehabilitation strategies to improve upper limb (UL) function in children with CP, the confidence in the validity of these studies’ evidence is still moderate to low. One limitation suggested is related to the type of outcomes used to measure improvement. Many are not sensitive enough to detect change (lack of responsiveness), are not age-related, and do not describe the movement quality. Movement quality concerns about movement performance or how well an activity is performed taking into reference normative data from typical populations. The assessment of movement quality in UL activities refers to the measurement of range of motion, hand trajectories, interjoint and intersegment coordination, muscle contraction patterns, and postural adjustments. The objective assessment of movement quality can be made by kinematic and kinetic analyses. The description of movement quality is important, because early brain injuries are more susceptible to ‘maladaptative’ plasticity, which might result in abnormal movement behaviors.

The primary objective of this prospective single subject research design study was to determine the effect of two rehabilitation strategies in UL movement quality: arm constraint and trunk restraint, in the context of a modified constraint induced therapy (mCIT) and a task-oriented intervention, respectively. The UL movement quality was measured by kinematic analysis of a functional reaching task: a self-feeding simulation. Overall, the kinematic variables investigated are related to hand trajectories, arm angles and trunk forward displacement. Two clinical outcomes measuring UL movement quality were also used, the QUEST for the mCIT study, and the Melbourne assessment for the task-oriented intervention study. Due to the high variability observed in movement patterns in young children with CP, the secondary objective of this study was to identify the most reliable kinematic variables that could be used in clinical trials to detect
change in UL movement quality. The results of the first study demonstrated that following mCIT, all three children improved QUEST scores and two children improved in at least one kinematic outcome variable. The results of the second study demonstrated that kinematic variables are highly reliable and can be used as outcome measures in clinical trials. For the third study, it was observed that children who practiced task-oriented intervention with TR strategy had better improvements in upper limb movement quality, and these improvements were maintained three months after the intervention. Furthermore, important movement compensations such as excessive trunk displacement were detected following UL practice in the mCIT and in task-oriented interventions without trunk restraint (TR) strategy.

Our results suggested that children with CP benefit from upper limb practice in a task-oriented intervention with TR strategy. Moreover, according to the results of this study, clearly, interventions aiming to improve UL function and activity in this population should address movement compensations. Further research to determine the responsiveness of kinematic variables and to better understand compensatory mechanisms in reaching and grasping activities in children with CP is needed. Such studies are necessary in order to provide details related to movement quality that can be incorporated in the development of pediatric outcomes measurements.
SOMMAIRE

Les enfants avec paralysie cérébrale (PC) constituent un groupe clinique fortement hétérogène, tant au point de vue de l’étiologie que des caractéristiques cliniques. La diversité des symptômes retrouvée parmi les syndromes de PC demeure un défi pour différentes branches de la recherche en santé. Malgré les efforts de recherche qui ont été déployés dans plusieurs études pour examiner les stratégies de réadaptation visant l’amélioration de la fonction du membre supérieur chez les enfants avec PC, il demeure que la confiance démontrée envers la validité de ces résultats est encore de modérée à faible. Une des limites notées dans la littérature est reliée au type de variables qui sont mesurées : ces variables ne seraient pas assez sensibles pour détecter des changements (manque de sensibilité des échelles de mesures) ou leurs valeurs ne seraient pas reliées à l’âge ou ces études ne décriraient pas la qualité du mouvement. La qualité du mouvement est centrée sur la performance du mouvement ou jusqu’à quel point l’activité est bien réalisée, en prenant comme référence les valeurs normales obtenues chez des populations typiques. L’évaluation de la qualité du mouvement pour des activités du membre supérieur se réfère aux mesures reliées aux amplitudes de mouvement, aux trajectoires de la main, à la coordination interarticulaire et intersegmentaire, aux patrons des contractions musculaires et aux ajustements posturaux. Cette évaluation objective de la qualité de mouvement peut être réalisée par l’intermédiaire d’analyses cinématiques ou cinétiques. La description de la qualité du mouvement est considérée primordiale, puisque des lésions cérébrales en bas âge sont plus susceptibles d’amener une plasticité « maladaptative » et donc d’engendrer des mouvements anormaux.

Le premier objectif de cette recherche prospective à protocoles individuels (single subject research design) est de déterminer les effets de 2 stratégies de réadaptation visant la qualité du mouvement du membre supérieur: la contrainte du bras (arm constraint) et la restriction du tronc (trunk restraint), réalisées respectivement dans le contexte d’une intervention par contrainte modifiée (modified constraint induced intervention, mCIT) et d’une intervention orientée
vers la tâche. La qualité du mouvement du membre supérieur a été mesurée par
l’analyse cinématique d’une tâche fonctionnelle d’atteinte consistant en une
simulation d’auto-alimentation. Globalement, les variables cinématiques mesurées
dans ces protocoles comprennent les trajectoires de la main, les angles articulaires
du bras et le déplacement du tronc vers l’avant. Deux échelles de mesure cliniques
rétroactives à la qualité du mouvement du bras ont aussi été utilisées : le QUEST (étude
mCIT) et le Melbourne assessment (intervention orientée vers la tâche). Étant
donné l’observation d’une grande variabilité au niveau des patrons de mouvement
chez les jeunes enfants avec PC, le second objectif de cette étude consistait à
identifier les variables cinématiques démontrant la meilleure fiabilité, avec
comme optique d’utiliser ces variables plus fiables dans des essais cliniques
visant à détecter des changements au niveau de la qualité de mouvement des
membres supérieurs. Les résultats de la première étude ont montré que les jeunes
enfants avec PC présentent une grande variabilité intra-individuelle et inter-
individuelle dans leurs patrons de mouvements d’atteinte. De plus, après avoir
suivi la thérapie mCIT, les trois enfants qui ont participé ont amélioré leur score
au QUEST et deux de ces enfants ont présenté une amélioration au niveau d’au
moins une variable de mesure cinématique. Les résultats de la seconde étude
démontrant que les variables cinématiques ont une forte fiabilité et peuvent être
utilisées en tant que mesures dans des essais cliniques. La troisième étude a mis
en évidence que les enfants qui ont participé à une intervention tâche-orientée
avec restriction du mouvement du tronc ont montré des améliorations plus
substantielles au niveau de la qualité de leurs mouvements avec le membre
supérieur, et ces améliorations ont été maintenues trois mois après la fin de
l’intervention. Par ailleurs, d’importantes compensations au niveau du
mouvement, telles qu’un déplacement du tronc excessif, ont été détectées chez les
enfants après leur participation à l’étude avec mCIT pour le membre supérieur et
dans le groupe impliqué dans l’intervention tâche-orientée sans restriction des
mouvements du tronc.
Ces résultats indiquent que les enfants avec paralysie cérébrale peuvent bénéficier d’une pratique des mouvements du membre supérieur dans le cadre d’une intervention tâche-orientée lorsque celle-ci implique la restriction des mouvements du tronc. En effet, les résultats démontrent clairement que les interventions visant l’amélioration de la fonction et de l’activité du membre supérieur dans cette population devraient prendre en compte les compensations motrices. Également, on note le besoin d’études supplémentaires pour cerner la sensibilité des mesures cinématiques et pour mieux comprendre les mécanismes de compensation survenant pendant les mouvements d’atteinte et de préhension chez les enfants avec PC. Ces études sont nécessaires afin de mieux détailler les éléments reliés à la qualité de mouvement qui pourraient être incorporés dans le développement de variables de mesures pédiatriques.
PREFACE

Thesis format

The format of this thesis is manuscript-based, and it was prepared in accordance to the McGill Graduate and Postdoctoral studies guidelines of thesis preparation. In the guidelines is estated that: “Candidates have the option of including, as part of the thesis, the text of one or more papers submitted, or to be submitted, for publication, or the clearly-duplicated text (not the reprints) of one or more published papers.” In agreement with these guidelines, this thesis contains three original papers, which have been submitted or will be submitted.

Chapter 1

Chapter 1 provides an overview of this thesis with a general introduction.

Chapter 2

Chapter 2 consists of a literature review composed of 10 sections with some aspects of cerebral palsy syndrome, knowledge about it, and the importance to rehabilitation of upper limb movement quality. The first four sections provide a review of the definition, etiology, clinical impairments and impact of upper limb deficits in function. The following three sections describe important aspects of movement quality in reaching in typically developed adults, children, and in children with CP. The last three sections address the evidence in rehabilitation interventions and strategies reported in the literature. Sections 9 and 10, describe the task-oriented intervention approach and the trunk restraint strategy used in the third manuscript. Section 11 is a brief summary of the important aspects in the literature review, section 12 the rationale, and section 13 the objectives and hypotheses of this study.
Chapter 3, 4, 5

These chapters are formed by the individual scientific papers, where the first and the third are preliminary proof of principle studies on the effects of two novel rehabilitation strategies on upper limb movement quality in children with CP, and the second estimates the reliability of kinematic variables, the main measures used in this study to determine movement quality. The first manuscript (chapter 3) consists of three case reports wherein the effect of modified constraint induced therapy on kinematic variables was characterized. The second manuscript (chapter 4) estimates the reliability of kinematic variables with a test-retest model. Finally, the third manuscript (chapter 5) determines the effect of task-oriented intervention with and without trunk restraint on upper limb movement quality in children with CP.

Chapter 6

This chapter presents a summary of the findings found in the previous three chapters. Following the summary is a discussion in which some additional materials not included in the manuscripts are presented, and a more intensive and extensive integration of the ideas exposed in the literature review is made.

Connecting texts

Preceeding each manuscript are connecting texts where the content of each manuscript is introduced and the integration between the three manuscripts is undertaken.

Appendix

This appendix contains all documents and ethics certificates that were necessary to conduct this study.

References

The references for chapters 1, 2, and 6 are compiled at the end of this thesis.
CONTRIBUTION OF AUTHORS


This manuscript will be submitted to the Pediatric Research journal. The text included in this thesis is exactly the same as the one submitted. The candidate of this thesis, myself, was the responsible for data collection, analysis, and manuscript preparation, under the supervision of Dr. Mindy Levin. The recruitment of the three children was performed by Dr. Celine Lamarre a physiatrist at Ste-Justine Hospital. The coordination of treatment delivery was done by Alain Bibeau, coordinator of the occupational therapist department at the Marie-Enfant Rehabilitation centre and Ste. Justine Hospital. All co-authors contributed to writing the above manuscript.


This manuscript was submitted in May 2009 to the journal Developmental Medicine and Child Neurology. In this study eleven children were recruited at Montreal, and two others in Ottawa. The clinical and kinematic data collected in Ottawa was in the majority collected by the candidate at Dr. Heidi Sveistrup’s motor control lab, located at University of Ottawa. The candidate was responsible for the recruitment, clinical and all kinematic data collection in Montreal, analysis and manuscript preparation, under the supervision of Dr. Mindy Levin and Dr. Patricia McKinley. All co-authors colaborated to interpretation of the findings and have reviewed this manuscript.

All of the data presented was gathered, analysed, and the manuscript was prepared by the candidate. The candidate was responsible for selecting, adapting, organizing and installing all equipment used in the treatment investigated in this manuscript, and in coordinating and training therapists in the five rehabilitation centers involved in this project. All the work was supervised by Dr. Mindy Levin and Dr. Patricia McKinley. The task-oriented intervention was elaborated by the candidate and Dr. Levin together with the expert opinion of Dr. Gisel who has years of experience with CP population, Dr. McKinley who provided her expertise in motor development and motor behavior analysis, and Dr. Sveistrup who contributed her expertise in motor development and contributed with the implementation and adaptation of the virtual reality games used as part of the treatment. The project was conceived by Drs. Levin, Sveistrup, Gisel and McKinley and funding was obtained for the project from CIHR in 2004. Thus, part of the data collection took place in the lab of Dr. Sveistrup. Dr. Nancy Mayo provided expert guidance for the single subject research design, and analyses of this manuscript. All co-authors have contributed to the interpretation of finding and have read this manuscript.

All the work contributed to this thesis as recruitment, data collection and analysis, proposal for scientific ethical approval in the five centers involved in this thesis, as well as coordination of treatment, instalation and maintenance of treatment equipments, and manuscript and thesis writing were carried out by the candidate. All the processes involved in this thesis were supervised and directed by Dr. Mindy Levin and Dr. Patricia McKinley.
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CHAPTER 1

GENERAL INTRODUCTION

Cerebral Palsy is a syndrome with multiple, etiologies, and symptoms (Baxter 2005). A unified definition of CP has been debated for years. It was just recently in 2004, that researchers and clinicians met and debated to establish a definition that incorporates all multiple aspects of CP (Bax et al. 2005). The vast heterogeneity found in CP syndromes makes early diagnosis and determination of exact numbers of incidence and prevalence difficult (Shevell and Bodensteiner 2004). For example in a cohort in Atlanta, Georgia, United States in the period of 1975-1991, was an increase in the prevalence of CP from 1.7 to 2.0 per 1000 was reported for survivor infants of normal weight, and no difference in prevalence in survivors with low weight (Winter et al. 2002). While in Alberta, Canada, the prevalence in CP in babies with low weight increased between 1992-1994, following a reversal decreased after 2001 to 1.9 per 1000 survivors (Robertson et al. 1998; Robertson et al. 2007). Prevalence in cerebral palsy in Quebec, Canada is still under investigation (Mathieu and Shevell 2003). Therefore, it is important to understand why research in the CP population has been a challenge for many health professionals.

The upper limb deficits that are present in children with CP have an impact on function and activity (Gordon and Duff 1999). Reaching, grasping and prehension are activities essential for daily life, that are performed with abnormal movement patterns by children with CP (Hirschfeld 2007). The fact that CP results from lesions in an immature brain, where high plasticity is present, demands further attention by researchers. It is necessary to be aware of the consequences that this high plasticity can bring to the flow of normal development and to skill acquisition (Gramsbergen 2001; Clowry 2007; Eyre 2007). Measurements of motor behavior using kinematic analysis, and recent advances in neurophysiological measurements are essential assessments to
distinguish between ‘good’ or ‘bad’ plasticity resulting from motor learning or skill acquisition (Alaverdashvili et al. 2008; Allred and Jones 2008).

Recent rehabilitation interventions based on casting and restraint strategies have been developed to improve UL function and activity in children with CP (Boyd et al. 2001; Charles and Gordon 2005). At present, the level of scientific supporting evidence in rehabilitation strategies to improve UL function and activity is low to moderate (Anttila et al. 2008). Researchers attribute this lack of evidence to the psychometric properties of the outcomes available and quality in methodology in the clinical trials.

This thesis presents the results of two studies that investigated the effect of two rehabilitation strategies: arm constraint and trunk-restraint, in the context of constraint induced therapy and task-oriented intervention approach. The reliability of kinematic variables to investigate UL movement quality was also estimated. By characterizing the effect of these interventions with detailed analysis of movement quality, we may be able to promote significant understanding of the mechanisms of motor behavioral change in this population that further can be used to improve pediatric outcome measurements and clinical practice.

This doctoral thesis will precede now with the sections of review of literature, rationale, and the objectives of the studies. These sections are followed by the presentation of three manuscripts, then a summary of the findings precedes the general discussion. Finally, the clinical relevance, significance, limitations and future research will be presented.
CHAPTER 2

LITERATURE REVIEW

2.1. Cerebral Palsy – The Definition: the beginning of the dilemma.

Reaching a consensus definition of the term Cerebral Palsy (CP) has been a challenge for many years (Shevell and Bodensteiner 2004). The multifactor etiology and diverse clinical manifestations of CP are the primary challenges in the effort to capture the essential meaning of this wide spectrum, ‘non-progressive syndrome’. The classical definition of CP is “an umbrella term covering a group of non-progressive, but often changing, motor impairment syndromes secondary to lesions or anomalies of the brain arising in the early stages of its development” (Mutch et al. 1992). This definition expresses the multidimensional aspect of CP while excluding progressive diseases. However, it is still ambiguous because of its incomplete description of clinical symptoms, its emphasis on motor impairments, imprecise development age, and its unclear etiology and disability impact. The need for better communication among researchers, clinicians and families, together with advances in developmental neurobiology, updated concepts about impairments, as well as functional status and participation (Grimby and Smedby 2001), necessitated a revision of the definition of CP.

An international workshop on the definition and classification of cerebral palsy was held in July 2004. Its goals were to revise and update the definition and classification of CP (Bax et al. 2005). The workshop emphasized that the new definition of CP was not an etiologic diagnosis, but rather a clinical descriptive term; Emphasis was placed on the concept that neurodevelopmental disabilities, such as CP, comprise several kinds of impairments which have an impact in different aspects of function. The goal of the new definition of CP was to aid in the diagnosis, management, and epidemiological aspects as well as to facilitate communication, research and the public health services.
The workshop agreed on the following definition: “Cerebral Palsy (CP) describes a group of disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, cognition, communication, perception, and/or behaviour, and/or by seizure disorder.” (Bax et al. 2005). This new definition includes important elements that facilitate diagnostic and clinical decisions: “group of disorders of the development of movement and posture” emphasizes that CP has an impact in development, which differs from other forms of brain lesions in adulthood, and that for the disorder be classified as CP; it should interfere with motor control. “Causing active limitation” implies that the disorder must cause difficulty in executing activities; otherwise, the disorder cannot be considered part of the CP disorder group. “Accompanied by disturbances of sensation, cognition, communication, perception, and/or behaviour, and/or by seizure disorder”, implies that often children with CP have other disorders and impairments besides movement and posture. The updated definition seems better able to describe the multidimensionality of the syndrome and hopefully it will be widely used, thus facilitating diagnosis, treatment and knowledge transfer for the best clinical practice within this population.

2.1.1. Multimodal etiologies of CP.

There are many causes of CP. Determining the exact etiology is not fundamentally important in order to treat motor impairment or associated disorders (Miller 2007). However, etiology is essential for early diagnosis, prognosis, incidence, as well as for understanding why the CP happens (Ferriero 1999; MacLennan 1999). The etiology can be separated according to when the brain lesion occurred (Miller 2007). Congenital etiologies consist of brain malformations in the embryonic stage. A deficit in neural tube formation in the embryonic stages is called encephalocele. Not all encephaloceles cause CP. Some
are related to Meckel’s syndrome, which is a general malformation not just involving the neural system, but organs and limbs. A segmental defect in the brain is called schizencephaly, which means that there is a fissure in the brain. Schizencephaly can cause minimal to severe disability and it is associated with spasticity, quadriplegia and mental retardation. Deficiency in the primary proliferation of the neuronal substrate of the brain is denominated microencephaly. Conversely, megalencephaly occurs when the brain is too large. This is related to cellular hyper proliferation. The neurons in the brain migrate towards the periphery during typical development. When this migration is disturbed, the results lead to lissencephaly, a decreased number of gyri in the child’s brain. The condition where there are too many gyri is polymicrogyria. Focal cortical dysplasia, or dysgenesis, is a defect that occurs during brain cortex formation. Focal cortical dysplasia is present in several forms of CP, and it is associated with seizure disorders. During typical development when synapses are continually being formed and remodeled, important seizure activity in the young brain can prevent synapses to remodel due to excitotoxic injury. This can also lead to CP (Miller 2007).

According to O’Shea (2002) and Miller (2007), prenatal and perinatal etiologies are related to prematurity and delivery problems which are major risk factors for developing brain hemorrhages. The bleeding can be of three types: intraventricular (IVH), inside of the ventricle; periventricular, or germinal matrix hemorrhage (GMH); or intraventricular-periventricular (PIVH). These hemorrhages can be graded according to severity from 1 to 4 (de Vries et al. 1993), in which 1 is when a GMH occurs, and 4 when a PVH and infarctions occur. The more severe the grade, the higher the risk of developing CP. A hemorrhage event may evolve into a periventricular leukomalacia (PVL) within 1 to 3 weeks after birth. Strokes involving the middle cerebral artery may take place in the pre-term or full-term infant and can be associated with the hemiplegic form of CP. Another vascular event that can occur during the delivery or neonatal stages is asphyxia or hypoxia, the latter is also known as hypoxic-ischemic encephalopathy (HIE). It has been reported that HIE in term infants can also lead
to CP (Nelson 2002). If the HIE is severe it can cause multicystic encephalomalacia and the prognosis for function is poor. Multicystic encephalomalacia is related to quadriplegia and severe mental retardation (Miller 2007).

The post-natal causes of CP are post-natal traumas, metabolic encephalopathy, infections and toxicities (Miller 2007). However, Shevell and Bodensteiner (2004) suggest that disorders which repeatedly insult the brain should be excluded from the etiology of CP. They do not agree that metabolic disorders, some specific vascular disorders (e.g. Moyamoya – a progressive disease of the cerebral vasculature with particular involvement of the Circle of Willis, or mitochondrial encephalomyopathy with lactic acidosis and strokes – MELAS – which is a progressive neurodegenerative disorder), and traumatic disorders as ‘shaken baby syndrome’ should be considered as causes of CP due to their progressive form.

An interesting study investigated the causes and frequency of CP over 10 years in a hospital and private clinic-based community (Shevell et al. 2003). The total number of patients in the database (1991-2001) was 6,616, 3.3% of whom (217) were identified as having CP symptoms. It was possible to identify the etiology in 178 of 217 (82%) cases. A single etiology was identified in 144 (66.4%) of the cases, and multiple etiologies were identified in 34 (15.6%) of the cases. The five most frequent diagnoses that contributed to the development of cerebral palsy were: periventricular leukomalacia, intrapartum asphyxia, cerebral dysgenesis, intracranial hemorrhage and vascular disorders.

“How to make a diagnostic of something that it is not one thing?” Ferriero (1999), in her comments in Current Opinions in Pediatrics, discusses the fact that most of the difficulties in diagnosing CP in its early stages are due to multiple etiologies. Although a severe case of CP can be easily diagnosed during the first months of life, in order to have a confirmed diagnosis in later CP cases it might be necessary to wait until 3 to 4 years of age (Dzienkowski et al. 1996).
2.1.2. CP Classification - Clustering a heterogeneous group.

The Cerebral Palsy syndrome has many ways by which it can be classified. Traditional classification is carried out according to impairment distribution on the area of the body, as for example, in monoplegia one limb is affected, in diplegia the lower limbs are affected, in hemiplegia the upper and lower limbs on one side of the body are affected, and in quadriplegia, all limbs are affected. The impairment distribution classification is followed by the type of tone, based on the most evident movement disorder resulting from brain lesions that comprise spastic, dyskinetic (athetoid), and ataxic types (Campbell et al. 2006). However, confusion can still occur in the classification of diplegia, which could be classified as a restricted involvement of lower limb. Others included in the diplegia group are children who, beside lower limb involvement, have an additional mild to moderate impairment in one of the upper limbs. The tone type is also a source of confusion for classification, especially when it is reported in clinical practice as ‘mixed’ without any definition. Rosenbaum and Stewart (2004) and Bax et al. (2005) have proposed that the International Classification of Functioning, Disability and Health (ICF) be used as a model to define, classify, treat and understand the needs of Children with CP. According to Bax et al. (2005) the classification of CP should be based on:

1) Motor disabilities: the nature and type of motor disorder, as well as the type of abnormal resting muscle tone. The type of tone or movement disorder - spasticity, dystonia, choreoathetosis, or ataxia - should still be used in the classification. However, the term ‘mixed,’ alone or without further elaboration, should be avoided.

2) Functional motor abilities: the ICF’s recent publication (WHO 2001) has emphasized the importance of evaluating the function consequences in several health states. CP should not be different. Functional consequences of motor impairments in the lower and upper limbs should be classified according to reliable
and objective scales. For the lower limbs the major activity impact is ambulation. In order to classify the impact of this function the Gross Motor Function Classification System – GMFCS – has been broadly used (Palisano et al. 1997). For the upper limbs, the most important consequence in function is the ability to handle and manipulate objects. In order to classify the severity of involvement in the upper limbs, the Manual Ability Classification System – MACS – was recently created (Eliasson et al. 2006b).

3) Anatomic distribution: the traditional classification according to the area of the body should remain. However, it was recommended that all body regions, such as trunk, each limb, and oropharyx be described in terms of impairment or posture deficits.

Research including the classification of CP radiographic findings, as well as causes and time of insult is considered important in order to complement an ideal classification system for this population.

The definition, etiology, the distinction between different clinical types (classification), incidence (case-finding/diagnosis), and changes in prevalence are problems affecting the epidemiological study of CP (Alberman et al. 1992). The exact numbers of incidence of CP from different studies do not completely agree. However, there is a remarkable similarity in the prevalence across the world: 1.6 per 1000 in China (Liu et al. 1999), 2.0 per 1000 from Atlanta (Winter et al. 2002), and 2.4 per 1000 in Sweden (Westboom et al. 2007). Progress in prenatal care, improvement in obstetrics conditions, and advancements in intensive neonatal care, made the incidence and prevalence of CP decrease over the years (Robertson et al. 2007). To date, however, there is no precise epidemiologic data about incidence or prevalence of CP in Quebec. It is fundamental to keep population-based registries in order to foster research to monitor changes in prevalence, and to plan care for children with CP. In 1998 an interdisciplinary
A group of specialists in rehabilitation (clinicians and researchers) initiated a task force to establish a registry of children with CP for the entire province of Quebec.


Knowledge of normal motor development is essential in order to plan a treatment for children with CP. Motor development refers to the acquisition of motor skills and age-dependent changes in perception, cognitive, neuromuscular and skeletal elements from infancy to adolescence. Karl Newell (1986) suggested that movement arises from the interactions of the individual with the environment in which the movement occurs, in the context of task-specific demands. If any of these three factors – the individual, the environment, or the task - change, the resultant movement will be modified. He called these the intrinsic and extrinsic factors of movement constraints, where the intrinsic factors are related with the individual himself and extrinsic factors with environment, experience and society. Thelen et al. (1987; Thelen and Smith 1994) have also suggested that movement patterns are the result of input from multiple systems that interact in dynamic ways to facilitate and constrain movement. These factors are both intrinsic to the infant, children or adolescent (e.g.: cognitive and perceptual ability, muscle size and strength, and/or biomechanical constraints and sensorimotor integrations) as well as extrinsic (e.g.: experience, task context and/or constraints).

These intrinsic and extrinsic factors have implications in the contemporary discussion about the hereditary or environmental elements involved in motor development. There are theories that sustain the idea of genetics in motor development: these promulgate the traditional notion that brain development is a genetically hard-wired process (Evans 1998). On the other hand, there are theories that emphasize the importance of environmental factors in motor development: that early stimulation and exposure assume an important role in many aspects of brain growth (Thompson and Nelson 2001). It is the interaction of these intrinsic and extrinsic factors that guide and shape development. For example, infants will acquire the ability to roll independently at different periods. A large overweight
infant may not roll until eight months but the first roll can be a mature one. A small infant may roll at two months, but use a primitive pattern of simple neck and arm extension (Case-Smith 1996).

The most important contribution of extrinsic factors to motor development is during periods of high-connectivity formation in the brain, also known as “wiring the brain” (Gabbard 2004). These periods are considered critical periods, or windows of opportunity. For basic motor skills, it has been suggested that there exists a window of opportunity from the prenatal stage to five years. Conversely, for fine motor skills, the window is open from the neonatal stage to about nine years. Finally, for second language learning, the period of opportunity closes around the age of ten years (Chugani 1998; Haris 1988). However, those ‘windows of opportunity’ can not be considered as inflexible for further learning or refinement of movement. For, example some aspects of motor development in reaching movements did not reach an established mature pattern in children up to 11 years (Schneiberg et al. 2002), which contradicts with the above reported opportunity period to acquire gross motor skill.

The knowledge regarding the role of hereditary and environmental factors in motor development naturally has an impact in rehabilitation treatment. Therapists should take advantage of windows of opportunity in order to obtain a greater effect during treatment. However, it is important to keep in mind that motor development is a process that is not exactly linear as previous theories based in the maturation of the central nervous system suggest (Gessel 1946; McGraw 1935, and Shirley 1931); motor development is rather highly variable, with peaks, periods of instabilities and even regressions (Savelsbergh et al. 1999).

2.2.1 Changes in the musculoskeletal system: shaping movement.

Besides the changes in the CNS during development, there are other changes resulting from musculoskeletal growth that also influence motor performance. Postnatal bone growth in length occurs at a secondary ossification center at the end of the shaft, termed the epiphyseal plate. Growth at the
Ossification centers cease at different times in various bones. Almost all epiphyseal plates are closed by age 18 or 19. In addition to the growth in length, bones also grow in circumference, which contributes to the additional weight of limb segments during development (Haywood and Getchell 2001).

Muscle cells grow during prenatal life by hyperplasia, which is an increase in the number of muscle cells, and by hypertrophy, which is an increase in muscle cell size. Hyperplasia continues only a short time after birth. Thus, most of the postnatal muscle growth is by hypertrophy. An adult muscle is composed of three types of fibers: type I (slow-twist) that are more resistant to fatigue and are used in endurance activities; type IIa and IIb (fast-twist) that are less resistant to fatigue and are used in intense and short-duration activities. At birth, muscles consist mostly of fast twitch units. After two years of postnatal life some units become slow twitch (Haywood and Getchell, 2001). In a study of 22 subjects aged 5 to 36 who had died accidentally, it was demonstrated that muscle cross-sectional area in the vastus lateralis more than doubled with age. There was also a 20% decline in the proportion of type I fibers between the ages of 5 and 20 years, suggesting that muscle fiber types become faster in the first 20 years of life (Lexel et al., 1992).

With the goal of analyzing if muscle maturation significantly contributes to dexterity, Lin et al. (1994) investigated the speed of alternating repetitive movements and correlated the findings with measured muscle twitch parameters (half relaxation time) in the ankle, metacarpo-phalangeal, and wrist joints in 38 children aged 3 to 11 years, and eight adults. They defined dexterity as the number of taps made by the hand or foot per second. They demonstrated that dexterity increased with age in all joints tested and the maximal speed of the joint tested showed a high curvilinear dependence of muscle half relaxation time (i.e. ankle tapping speed decreased with the increase of half relaxation time of the soleus muscle). They suggested that in addition to neuronal maturation, the factor responsible for the maturation of dexterity is the muscle itself as some of the mechanisms by which muscle dynamics change with age may reflect changes in the calcium re-uptake mechanism of the sarcoplasmic reticulum, which is known
to control muscle relaxation (Lin et al. 1994). Changes in inertial and physical properties of muscle such as viscoelasticity, resistance due to fiber type composition and muscle contractile properties also influence movement outcome. These properties are in continuous change during motor development obliging the system to adapt to them (Connolly et al. 1997).

2.2.2. Changes in the nervous system during initial stages of development: shaping movement.

The central nervous system (CNS) early developmental changes can be described in six dynamic events: cell proliferation, migration, integration, differentiation, myelination, and cell death (Gabbard 2004). The cell proliferation process occurs when the number of neurons in the brain increases. Neurons first appear in the brain during the second prenatal month, and the proliferation process has ceased by birth. The third trimester of gestation is a period of rapid brain growth and development. This period is also called the brain growth spurt, and continues at least until the fourth year. In parallel with the cell proliferation period, cells migrate to their final location in the CNS, and their axons and dendrites start to specialize and initialize synaptic connections.

There are different factors that determine cell migration and the growth of axonal connections. Cell migration appears to be directed by glial cells and interactions with cell adhesion molecules. At least four different mechanisms serve to guide axon path-finding: contact attraction, contact repulsion, chemotraction, and chemo-repulsion. In contrast to the complexity of cues driving axon outgrowth, there seems to be two mechanisms driving the early outgrowth of pyramidal neuron dendrites. First, dendritic growth is due to genetically determined, activity-independent signals. Second, incoming axon processes can induce dendrites to form. Dendritic differentiation and elaboration may be dependent of afferent input, as observed in experiment in rats, where some of them were deprived of sensory experience and the other group was placed in an
enriched environment. The rats in the enriched environment had more dendritic differentiation (Diamond et al. 1976; Webb and Monk 2001).

The ‘wiring’ event during neural development involves the migration of neural structures toward their target destination. The differentiation phase occurs when neural structures become more specialized and motor control improves to more precise and complex forms of motor behavior. Neural cell differentiation in the brain is only possible with synaptogenesis. Many researchers have proposed that there is initially an overproduction and redundancy in synaptic connections, followed by a period of reduction (Huttenlocher and Dabholkar 1997; Goldman-Rakic 1987). Originally, this pattern of sculpting may rely on endogenous, spontaneous neural activity. However, with maturation, increases in sensory input from the environment promote more influence in neural patterning. Stabilized synapses may represent coordinated activity at presynaptic and postsynaptic sites (Schlaggar et al. 1993), while synapse stabilization may occur through the local release of neurotrophins and brain-derived neurotrophic factors. It is possible that the circuits that stabilize and persist may be those that benefit from the great amount of activity, while those that regress may be lacking activity (Hadders-Algra, 2000). This process of synaptic elimination may play a crucial role in the development of an optimal function in the brain.

The development and arrival of cortical-spinal input in the spinal cord is essential in the development and refinement of spinal cord circuitry. According to Clowry et al. (2007) the human cortical-spinal tract decussates around the 11th to 13th post-conception week (PCW). The connectivity with cervical spinal circuitry begins around the 25th to 35th PCW, while the process of myelination starts around the 36th PCW and continues until 6 years. However, the development process of myelination can continue through adolescence to adulthood, and into old age, in certain brain areas (Huttenlocher and Dabholkar, 1997). In some areas in the brain, up to 70% of cells die during development. The survival and death of a cell is also linked with activity or experience. Synapse elimination can also occurs at the neuromuscular junction. Redfern (1970) has studied the innervation of skeletal muscle cells in the rodent. He found that at birth essentially every
muscle in the rodent was innervated by two or more motor axons, but around the first 3-4 weeks of postnatal life each fiber became innervated by one axon. In addition, if one of the innervating axons innervated any muscle fiber and was subsequently stimulated, it led to a selective loss of other axonal inputs to that muscle. Similar results, but with a different technique were obtained by Callaway et al. (1987), where they have found that when the activity of a nerve innervating a muscle is blocked with tetrodotoxin, the motor units connected with this inactive nerve are enlarged.

The multiple interactions involved in the nervous system’s development are complex, and disturbing any process during development can lead to diverse abnormalities. According to Thelen (2000) and Eldeman (1992), cell division and migration are the driving intrinsic factors of development, while connection, differentiation, and cell death are regulatory processes. Knowledge about the changes in the CNS during typical development (TD) can be used to detect abnormalities in the development of children with CP (Kulak et al. 2006). Nowadays with imaging and neurophysiological techniques, it is possible to compare intrinsic neural mechanisms responsible to voluntary movements in a TD brain with the ones in the injured brain.

The consequences of early brain damage are completely different from that of later ages (Gramsbergen 2001). Moreover, in adults, impairments usually appear almost immediately after the brain injury. However, in early brain injury there is a delay in the appearance of impairments, that might be due to the fact that the brain circuitries are not yet established. Additionally, it is difficult to relate the severity of impairments to the size of lesions in early brain damage. A lesion in an immature brain might also result in aberrant ipsilateral corticospinal projection (Thickbroom et al. 2001). These abnormal ipsilateral connections may not replace the function of contralateral tracts in organizing the segmental mechanisms in the spinal cord (Clowry 2007). Previous studies demonstrated that in neonatal rats with unilateral brain lesions, the ipsilateral cortex may replace the function of the impaired corticospinal tract by the formation of indirect pathways via the ipsilateral red nucleus (Gramsbergen 2001). Similar findings with
transcranial magnetic stimulation (TMS) and functional magnetic resonance imaging (fMRI) in children with hemiplegic CP (Thickbroom et al. 2001) have demonstrated in the affected hand an abnormal ipsilateral hemispheric projection with shorter latencies than the contralateral hemisphere of the brain. Furthermore, passive and active movements activate the contralateral hemisphere, suggesting an inter-hemispheric dissociation between afferent kinesthetic inputs and efferent corticomotor output. The authors also suggested that size, location of lesion, and the level of maturity of the system at the time of injury might be considered as determinants factors in the inter-hemispheric reorganization of cortical motor control.

As discussed above sensory information is essential for normal motor development. In some studies has suggested that children with CP have problems integrating the sensory information for planning their movements (Thickbroom et al. 2001). Therefore, it is expected that children with CP have larger differences in motor development among themselves and when compared to typical developing children.

2.3. Clinical features of CP: Making the diagnosis

There is a general notion that brain lesions at the perinatal age have less severe consequences on motor behavior than similar lesions occurring at adulthood. This notion is supported by findings in experimental research in monkeys (Gramsbergen 2001). However, these findings refer to brain lesions in the infant stage. With advances in neurophysiological techniques and careful observation of behaviour it has been demonstrated that newly formed connections or plasticity are not always beneficial to motor function (Alaverdashvili et al. 2008). The term plasticity is defined as the capacity of modification in neuronal interactions by changing neural circuitry or synaptic efficacy. Plasticity is often estimated as beneficial after brain damage. However, the maturation process in the CNS - myelinization, cell death, synaptogenesis, dendritic arborization, and brain ‘wiring’ - has not been completed during the perinatal period. A brain lesion
prior to or during the critical periods of development affects motor behavior differently than a lesion occurring at another time (Gramsbergen 2001).

The pattern of motor behavior due to brain damage depends on the maturity of the brain at the time of the brain insult. Moreover, different neural circuitry appears to control motor behavior at different stages of development. Certain motor behaviors which may be unaffected at the time of a lesion may display deficits following further development (Goldman and Galkin 1978; Leonard and Goldberger 1987). Therefore, the consequences to motor behavior from identical types of lesions at different epochs might vary, adding a crucial heterogeneous quality to the clinical features observed in children with CP.

Neonatal clinical assessments in children suspected of having CP are generally carried out by investigating muscle reflexes, tone, and elicited reactions (Olney and Wright 2006). However, as discussed before, there is much discussion about how early CP might be identified. Often, as discussed in the etiology section, the accuracy of a CP diagnosis at 2 years of age is not reliable if the involvement is mild (Kitchen et al. 1987). In contrast, Olney and Wright (2006) suggested that the availability of discriminative tests and scales allow an experienced therapist or pediatrician accurately to diagnose CP in all but the mildest cases by the age of 6 months. The authors reported that the Movement Assessment of Infants (MAI) has been demonstrated to have more sensitivity to identify children with CP at 4 months of age than the Bayley motor scale (Harris 1987). However, the Bayley scale has a higher rate of specificity than the MAI, as it is capable of classifying spastic diplegia and quadriplegia (Weisglas-Kuperus et al. 1993). The Alberta Infant Motor Scale (AIMS) is able to identify severe motor development delay in all children at 6 months of age, but it is not able to detect development delay at 3 months (Darrah et al. 1998). The Test of Infant Motor Performance (TIMP) shows promise of very early detection of CP and differentiation between infants with brain lesions and those born with less than 30 weeks of gestational age (Campbell et al. 2002). Imaging studies are still infrequently used to make diagnosis or prognosis. Usually, they are only required to rule out other treatable disorders such as tumors or hydrocephalus (Miller
In addition, Miller’s opinion is that imaging exams can help to screen for infants at risk for later disability but they are not helpful in predicting the course or the type of disorder (Miller 2007). Fundamentally, because the determination of diagnosis in early stages is still an area that needs further research, it remains a question of how much rehabilitation treatment could be accomplished after the diagnosis is made, as well as the impact that early treatment could have in preventing major disabilities in these children.

2.3.1. Clinical features of CP: Impairments.

The clinical features of CP can be classified as single or multiple system impairments (Olney and Wright 2006). Single system impairments are defined as secondary disorders of the neuromusculoskeletal system as alterations in muscular tone, hyperactive reflexes, weakness, misalignments or skeletal deformities. Multiple system impairments include central or primary disorders that also include the neuromuscular system but consist of high order functions such as motor planning and control. Multiple system impairments include poor selective control of muscular activity, abnormal patterns of movement, anticipatory postural regulations, and decreased ability for motor learning.

The term tone is used to describe the normal passive resistance of muscles to movement (Shepherd 1995). Hypotonus is expressed by extreme flaccidity when the infant is passively mobilized or by the infant’s inability to generate enough muscle contraction to produce voluntary movements against gravity. The clinical aspect of a hypotonic infant while lying in supine is the presence of externally rotated, lightly flexed and abducted legs. While in prone position, one can note the absence of protective side turning of the head, risking suffocation. When pulled up to sitting or standing position, the infant may be unable to hold her head up, or her lower limbs can collapse into flexion (Shepherd 1995). Hypotonus is often a transient clinical feature present in early stages of development. In later stages of development it could be reclassified as spasticity.
or athetosis and dystonia. The athetoidal form of CP is characterized by tonus fluctuation and involuntary movements (Olney and Wright 2006).

Spasticity is the most common impairment in children with CP (Richards and Malouin, 1992). It has been suggested that spasticity should be considered a component of Upper Motor Neuron Syndrome (UMNS), which also includes a decreased capacity to activate muscles in order to produce voluntary movement (weakness or paresis), abnormal coactivation of antagonists, as well as spasms and abnormal tonic stretch reflex responses (Richards and Malouin, 1992). It is important to note that the UMNS in CP is not identical to that observed after CNS lesions in adults, since in CP lesions occur in an immature CNS. It has been suggested that both supraspinal and interneuronal mechanisms are responsible for spasticity in CP (Young 1994). Three major peripheral pathophysiological mechanisms can be attributed to the cause of spasticity in CP: 1. reduced reciprocal inhibition of antagonist motoneuron pools by Ia afferents; 2. decreased presynaptic inhibition of Ia afferents; 3. decreased nonreciprocal inhibition by Ib afferents (Olney and Wright 2006). Shepherd (1995) has suggested that children with CP might be divided in two groups according to the severity of spasticity detected in early stages. She suggested that children whose spasticity is detected early belong to a group characterized by severely affected brain dysfunction in the cerebral cortex, midbrain and spinal cord. The other group, whose spasticity is not present in early developmental stages, consists of those cases where the hypertonus gradually develops due to changes in the functional properties of the muscle and neural improvement process, especially at the spinal level. Children who have severe impairments and who experienced important perinatal brain dysfunction are usually classified as dyskinetic CP (Himmelmann et al. 2007). The dyskinetic form of CP is found in cases of injury to the basal ganglia occurring in perinatal, neonatal or late gestation in the term or near term infant. These are periods when the basal ganglia are more vulnerable because of high metabolic demands. Interestingly, in contrast to the overall decrease in prevalence in CP due to improvements in obstetric and neonatal care, an epidemiological study in Sweden has reported that the dyskinetic form of CP has not decreased
since 1960 (Himmelmann et al. 2007). The dyskinetic type of CP are sub-grouped into choreoathetotic and dystonic forms. Children classified with dyskinetic CP have primitive reflex patterns: involuntary movement that can occur both at rest and during movement. Muscle tone can vary, but is often misclassified as spastic CP (Shepherd 1995). According to the Surveillance of Cerebral Palsy in Europe (SCPE), the dyskinetic form of CP represents 6.6% of the population (McManus et al. 2006). Another form of CP consisting of severe impairments is the ataxic form. The ataxic form, when found in children diagnosed with CP, is characterized by dysmetric movements, as well as uncontrolled movement velocity, force, range and direction, suggesting cerebellum involvement (Shepherd 1995). The incidence of ataxic form of CP in Europe (1980 -1996) was 4.1%.

Muscular weakness is an important impairment in children with CP (Damiano et al. 2002). It has been observed that 50% of the variance in walking speed can be explained by muscle weakness in children with the hemiplegic and diplegic forms of CP. The weakness of children with CP can be attributed to the lack of activity observed in this population, the decrease of central input to the muscle, any changes in the elastic properties of the muscle, and by spasticity. Exercise programs to improve strength in children with CP have been proven to have an impact in walking and prehension function (Anttila et al. 2008), social and participatory activities, and self-perception (Damiano and Abel 1998). Contractures and skeletal deformities develop when active control or muscle length can not be maintained in children. Skeletal deformity is more found in children with severe cognitive and motor impairments (Shepherd 1995).

Multiple system impairments present in children with CP can affect posture and voluntary movements. Children with CP have problems maintaining their balance during standing and walking functions (Woollacott and Shumway-Cook 2005). The reactive balance, which is the ability to regain balance from an unexpected perturbation, is impaired in children with CP. According to Woollacott and Shumway-Cook (2005), children with CP do not increase muscle responses when balance threats increase in magnitude, a typical finding in
typically developed (TD) children. Moreover, during gait analysis in children with CP, it has been observed that the presence of crouched posture, impaired muscle activation, loss of selectivity in muscular patterns (co-contraction), and spasticity, are underlying problems that are also common to balance deficits. The function of gait can be divided in two phases: stability (maintaining balance) and progression (moving forward) factors. Therefore, the authors suggested that the dysfunction in gait observed in children with CP reflects their underlying instability or balance problems.

Nashner et al. (1983) observed that the response to postural perturbations in children with CP differs according to the type of impairment distribution. For example, children with diplegic and hemiplegic CP experienced a reversal of the normal bottom–up muscle recruitment seen in TD children, while children with the ataxic type of CP had longer latencies of muscle responses, despite the order of muscle recruitment being similar to TD children. Another study investigated postural control while sitting in ten children with mild to severe forms of CP (Brogren et al. 1998). There it was found that during forward translations children with CP have a stereotyped and non-variable activation of all ventral muscles, a top-down recruitment, and an excessive degree of antagonist co-contraction. However, during reaching tasks children with CP had more variable postural muscle activity, an absence of coactivation in antagonist muscles, and abnormal top-down or cranial caudal muscle recruitment as compared to the age-matched control group (van der Heide et al. 2004).

It has been suggested that children with CP have motor learning deficits (Olney and Wright 2006). Motor learning can be defined as a new acquisition or improvement of function, or modification of movement. It is the result of a complex interaction of cognition, perception and action (Shumway-Cook and Woollacott 2001). According to Newell (1991), in order to learn a new motor skill children have to coordinate perception and action with the environment or task constraints. Children with CP have motor and sensory impairments that hamper their ability to perceive and act as TD children (Kulak et al. 2006), and these impairments may cause difficulty in the way that children with CP learn or
acquire movement patterns. No study to date has investigated the motor learning of children with CP compared to TD children. While many studies have investigated motor learning in adults following brain injury (Proteau et al. 1994; Plautz et al. 2000; van Dijk et al. 2005), few have addressed the cognitive aspects of learning in children with CP (Wann and Turnbull 1993; Thorpe and Valvano 2002). Thorpe and Valvano (2002) investigated the motor learning inherent in moving a pedal of a therapeutic exercise vehicle. They randomized the children into three different learning strategy categories: 1. no augmented information; 2. knowledge of performance; and 3. knowledge of performance enhanced by a cognitive strategy. Though all subjects improved their performance, the majority of subjects performed better when the learning strategy used was knowledge of performance enhanced by a cognitive strategy.

In order to evaluate multiple system impairments in children with CP, it is necessary to investigate different types of variables. This follows as a result of the fact that the consequences of these impairments affect many numbers of functions. Usually, multiple system impairments are investigated with kinematic, kinetic, electromyographic (EMG) and, more recently, with neurophysiological techniques such as transcranial magnetic stimulation (TMS) and functional magnetic resonance imaging (fMRI). Kulak et al. (2006) have also suggested the use of quantitative electroencephalography (EEG) and proton magnetic resonance spectroscopy (H MRS) to determine neuphysiological and neuranatomical organization in children with CP.

2.4. Impact of CP in upper limb function.

The importance of upper limb (UL) function for the many activities of daily living is well known and accepted by all clinicians and researchers who work with movement. Without arm movement, hand mobility is limited, and, many fine-motor skills would be difficult or impossible (e.g. painting and writing). The major activities affected by the impairment in UL function are reaching, grasping and object manipulation. In addition, UL function also plays a
role in gross motor skills such as crawling, walking, and maintaining balance (Shumway-Cook and Woollacott 2001). Impairments in UL function are often barriers to several activities and social interaction for children with CP (Shikako-Thomas et al. 2008; Wright et al. 2008).

In a population-based study carried out in Sweden, Arner et al. (2008), have investigated hand function in children with CP using the Manual Ability Classification System (MACS), the House functional classification system, and the Zancolli classification. In 367 children, 64% (236) were classified at level I and II of MACS, which implies that the children were independent regarding age-relevant activities using both hands. For the House classification assessment only 55% were stage 7 or 8, which is considered an independent use of one of the hands. In the Zancolli assessment, which is normally used to grade the tone levels in the wrist and fingers, 69% (252) children did not have flexion spasticity in both hands, 86 children (23%) had moderate levels of spasticity in one or both hands and only 8 children (2%) had severe spasticity with no activity of wrist or finger extension. The authors reported that 21 children were not scored with the Zancolli classification assessment. This study also determined the distribution of hand function in relation to the type of CP. For all three assessments, the less impaired levels of classification were for children with hemiplegic CP and the most severe cases of hand function were with quadriplegic or tetraplegic CP.

Hanna et al. (2003) have investigated the development of hand function and UL movements in children with CP of ages approximately 1 to 5 years (16 to 70 months), using the Peabody Developmental Motor Scales (PDMS; Folio and Fewel 1983) and the Quality of Upper Extremity Skills Tests (QUEST; DeMatteo et al. 1992). The authors used a hierarchical linear model (Growth Curves) to estimate the development of hand function and upper limb movements in terms of average change over time of the PDMS and QUEST results, and to estimate the individuals' difference in the pattern of change. The results were extremely interesting: 1. Children with CP had high individual differences in the parameters of growth curve in both PDMS and QUEST scales. However, regarding the PDMS score, it was estimated that young children with CP would increase their
scores over time, but with a lower rate of change than TD children. The PDMS growth curve was also able to differentiate the abilities and estimated difference rates of development in children with mild, moderate and severe impairments. Children with mild impairments, and classified as hemiplegic CP, were estimated as having a comparable developmental rate in the PDMS scale as TD children. In contrast, quadriplegic children with all levels of severity had lower rates of developmental change on the PDMS scale when compared to hemiplegic children.

2. For the QUEST scores, the individual differences were substantial in all stages. Furthermore, the average function of UL movements estimated with the QUEST scores had a tendency to peak and decline after 46 months of age. The authors concluded that the PDMS scale and UL movements estimated with the QUEST scale estimated hand function development differently in children with CP.

One of reasons provided by the authors to explain the differences in the estimated curve growth between PDMS and QUEST was related to the type of task evaluated for each scale. The PDMS scale assesses more activities, such as drawing and cutting shapes, while QUEST assesses more impairments, such as wrist extension and dissociation of movements. In addition, it could be that the observed decline in the QUEST during development is associated with progression of impairments. As the child grows, impairments are more susceptible to muscle shortening, and repetitive movement compensations. Often development can be followed by an increase in spasticity or the appearance of contractures. The initiative shown by the study in estimating the development of hand function in children with CP is appreciated, since there exists a lack of studies documenting the development of hand and UL movements in this population. However, a possible explanation for the difference in hand function development and arm movement which remains to be investigated is the relation of hand function development and the type of impairment. In order to estimate development of hand function in children with CP, perhaps the model should include more predictors such as sensory tests, ranges of motion, level of UL activity (MACS), or gross motor mobility (GMFCS). Nevertheless, the data in
this study was retrospective from a sample collected for a clinical trial (Law et al. 1997). This would explain why the authors could not have used updated measurements of hand ability classification and mobility since the MACS was created around 2006 and the Gross Motor Function Classification System only became available after 1997.

In another small study Eliasson et al. (2006a) have investigated the development of manual dexterity using the Jebsen-Taylor test of hand function, force timing and amplitude control during a precision grasping task in children with CP. The authors made two assessments in a group of ten children with CP. The second assessment was 13 years after the first assessment. In the first assessment, children were age 6 – 8 years. The authors found that after 13 years the Jebsen-Taylor test scores improved, although they mentioned that two subjects did not perform the simulation of the eating task in the first and second tests. They also reported that the time of finger contact to lift the object decreased and that the loading phase was longer in the second assessment, which expressed improvement in the prehension task with age. However, they found that the majority of subjects did not develop a force coordination pattern, but instead had a maladaptive pattern considered abnormal for their age.

Based on the study findings discussed above, it is evident that UL movements and hand function are linked, and that CP has an impact on UL and hand function. Furthermore, little is known about how children with CP develop and how development influences UL function and activity. It is crucial to understand how children with CP develop, in order to provide effective treatment interventions and to promote efficient strategies that improve UL and hand function.

2.5. Insights into reaching movement: hand trajectory formation.

The capacity to perform hand interactions with objects (fine manipulation), or while walking, maintaining stability, reaching or grasping (gross movements) using UL movements is only possible when the
somatosensory and motor systems are functioning in collaboration. Studies concerning the reaching to a target in humans and nonhuman primates have documented two distinct phases: the transport and then grasping (Jeannerod et al. 1984). In addition, Pigeon et al. (1998, 2000) have suggested that reaching movements are also composed of several units of coordination or synergies. The first is the transport synergy which is aimed at changing the arm configuration according to the desired direction and extent of movement. The second is an arm-trunk synergy, used when trunk movement is also required to increase the reaching distance. The reaching distance determines the relative contribution of arm and trunk movement during the transportation of the hand to the target. The third is the grasping synergy whose functional goal is to prepare the hand aperture, shape and orientation for grasping an object.

In order to reach to an object, it is first necessary to locate the object in space. The cortical control of visually guided reaching involves many structures, such as the primary motor cortex, the pre-motor cortex, the supplementary motor area, the cingulate motor area, the primary somatosensory cortex, and the posterior parietal areas, as shown by studies on the distribution of reaching movement-related cell activity in the cerebral cortex of the macaque monkey (Kalaska et al. 1997). The control of reaching also involves sub-cortical structures like the basal ganglia and the cerebellum. The neural systems that control visually-guided reaching movements are the results of a process of integration of different sensory modalities that are used to build an internal representation of space. This spatial representation could be formed by different modalities such as vision, somatosensation, audition, and vestibular sensation (Andersen 1997). Many studies support the idea that spatial representations of limb position, target locations, and potential motor actions are combined in the posterior parietal cortex. Interesting findings were found in the superior parietal cortex (SPL) area 5 of Brodmann, which for many years was considered only a somatosensory region. Results of some studies have shown that neurons in this area respond during active arm movement towards a target more than during random arm movements. It could be that this area is responsible for the direction of joint movements, and
also that it can supply the frontal motor lobe and pre-motor areas not only with proprioceptive information but also with visual input (Kalaska 1996). Although reaching is commonly followed by grasping, anatomical evidence and neurophysiological recordings have demonstrated that separate but parallel parieto-premotor channels mediate visuo-motor transformations for reaching and grasping (Kandel et al. 2000).

Any damage to the pyramidal tract can result in impairments of fine finger control, and thus can impair the manipulation of objects. Damage to the extrapyramidal tract results in impairment of gross arm movements, thus impairment in hand transport. In addition, developmentally, the pyramidal tract also matures later than the extrapyramidal tract (Rosenbaum 1991). Some studies argue that in spite of their independence in neuromotor control, reaching and grasping are temporally and spatially coordinated (Gentilucci et al. 1992; Hoff and Arbib 1993). Others have said that the coordination between the two components involves more than a temporal coupling and a higher order control system is responsible for their integration (Jakobson and Goodale 1991; Marteniuk et al. 1990). Some findings that support the existence of coordination between reaching and grasping refer to the dependence on speed for the maximum separation between the index finger and the thumb when the hand is brought towards the object. The fingers widen increasingly when the hand travels at higher speeds. Another kind of dependence between transport and grasp concerns aperture timing between index finger and thumb that coincides with the deceleration inherent in the approach phase or the transport phase (Jeannerod et al. 1984).

It has been suggested that a reaching movement is planned in hand or endpoint coordinates (Georgopoulos et al., 1986; Flash and Hogan, 1985; Flanagan et al., 1993, Gordon et al., 1994). According to Rosenbaum (1991), if the motor system, instead of selecting a direct path of the hand to a target, rather selects a convenient set of muscle torques, one would expect simple patterns of joint angle and complex hand patterns. In contrast, if hand path is planned, the opposite will occur. Rosenbaum (1991) suggested that the motor system may plan
the movement with respect to joint space using the intrinsic coordinates of the body, or with respect to hand space using the extrinsic coordinates of the external environment. Morasso (1981) analysed hand trajectories in healthy adults when they pointed to targets. He found that subjects’ hands tend to move in a straight line, and that their joints demonstrated complex angular changes. Another study (Abend et al., 1982) reported that even when subjects were asked to draw curved lines, hand trajectories were composed of a series of straight line segments. These studies support the view that the motor system plans reaching in the hand extrinsic coordinates. However, Soechting and Lacquaniti (1981) found some invariant relationships among the joints during a pointing task, such as the same time-to-peak velocities of elbow and shoulder, as well as equality in the ratios of peak velocity and radial distance that the joints moved. This would suggest that planning could be done in intrinsic coordinates. Rogosky and Rosenbaum (2000) questioned whether space-based motor planning occurs at higher, equal or lower levels of the control system than joint-based motor planning. They based their conclusions on the following prediction: if spatial planning can be learned more quickly than joint planning, then this can be taken to suggest that spatial planning occurs at a higher level of control than joint planning. In contrast, if joint planning can be learned more quickly than spatial planning, that joint planning occurs at a higher level than spatial planning. They asked 32 healthy subjects to reach towards a target while following a visual display, which was distorted with respect to spatial hand displacement (space-based distortion) or with respect to joint angle displacements (joint-based distortion). They found that subjects adapted more easily to space-based distortion. Thus, the result supports the view that space-based planning occurs at a higher level than joint-based planning.

Latash (1993) agreed with the majority of the studies cited above when he wrote that the workpoint, the most important point for executing a task (i.e.: in the case of a reach-to-grasp movement, the workpoint is the fingers or the palm of the hand), is the focus of concern of the CNS, because its trajectory is vital for executing the task. However, in his opinion, even the trajectory of the workpoint is not a variable that the CNS uses to control the movement. He points out that if
an unexpected perturbation occurs during a movement, the trajectory of all points including the workpoint will change immediately while the central command presumably remains the same, until motor corrections are introduced. This indicates that the central command should stay invariant for some time independently of events in the periphery (Feldman, 1998). Thus, the invariant central command may be associated with a control parameter or variable: the lambda (λ) model as suggested by Feldman (1986), and others (Latash, 1993, Feldman and Levin, 1995). For Feldman and Levin (1995), kinematic and electromyographic patterns are not programmed but are instead emergent properties from the interaction among the system. They proposed that the CNS uses control variables (CVs) to produce voluntary movement. CVs are specified by the nervous system independently of current external conditions. Thus, biomechanical variables are not CVs but are influenced by them. Frames of reference or systems of coordinates are organized by the CNS and movement is produced by shifting the frames in space. The factors that define the frame of reference are derived from the Equilibrium Point hypotheses (EP) or lambda (λ) model. Thus, alpha (α) motoneuron threshold properties, proprioceptive feedback and components of the tonic stretch reflex are all factors that define the frame of reference. The neuromuscular system is a particularly dynamic system in which forces are position-dependent. Specifically, in order to bring such a system from one point of equilibrium (a combination of steady state values of position and forces) to another equilibrium point, the controller must change parameters that are independent of state variables (state variables are forces, velocity, and other derivates of motion law). According to the EP hypothesis, the goal is identified by the nervous system in the environment using afferent information while trajectory is controlled by the appropriate thresholds (λs) (Feldman 2008).

An alternative view of movement control is based on internal models. Imamizu et al. (1995) have investigated how the human motor control system solves the problem of trajectory planning, inverse kinematics, and inverse dynamics. They proposed two solutions: feedback control and feedforward control. The authors suggest that motor control is based on internal models, also
called the force control hypothesis. The force control hypothesis states that: 1. the central motor command is the specification of forces; 2. their values are derived using an internal model of inverse dynamics; and 3. predictive control mechanisms are based on forward and inverse internal models. In this formulation, desired movement trajectories are planned first in terms of spatial coordinates and their derivatives and then transformed into required forces and torques. In order to compute torques, the system uses an internal representation of dynamical equations of motion of the body interacting with the environment. This computation (realized by a set of neuronal networks) is called inverse dynamics, because it generates value of torques based on kinematics and thus inverts the input/output relationships inherent in actual (direct) dynamic laws. In the inverse-dynamics, torques cause changes in kinematics rather than the other way around (Hogan 1990).

To answer how hand trajectory is planned by the nervous system is not the goal of this review. However, understanding the different ways that hand trajectory can be investigated and how trajectories can deviate from a normal performance is important to determine underlying mechanisms of impairment in UL movements in children with CP. In addition, because hand trajectory formation involves the interaction of multiple systems, substantial information with corresponding analysis can be obtained in order to understand the movement quality performed during reaching and grasping tasks. This study is in the frame of equilibrium point hypothesis, since the variables of movement quality analysed were related to position and its derivatives.

2.5.1. Insights into reaching movement: arm and trunk coordination.

Most of the studies of reach-to-grasp movement consider reaches that are only made by the arm. However, in many daily situations, reaching occurs in which objects are placed beyond the limits of the extension of the arm (Wang and Stelmach 1998). In those situations the motor control system needs to coordinate arm and trunk movement. It has been suggested that arm and trunk motions are
governed by different neuromotor synergies (Ma and Feldman 1995; Kaminski et al. 1995; Saling et al. 1996; Wang and Stelmach 1998). Ma and Feldman (1995) demonstrated that in reaching tasks, the addition of trunk motion did not affect endpoint trajectory. They and others (Pigeon et al. 1999) also suggested that reaching in the limits of arm’s length involves two synergies: a reaching synergy that consists of moving the arm joints to displace the hand towards the object, and a second synergy that consists of moving the trunk and arm joints without affecting the position of the endpoint. Adamovich et al. (2001) also studied reaching movement involving the trunk. Subjects had to make fast arm movements without corrections to the targets while the trunk was either free to move or restrained. They found minimal changes in the hand trajectories and velocity profiles of the endpoint in response to trunk restraint, and these few changes were seen only late in the movement. Interestingly, the pattern of interjoint coordination substantially changed during trunk arrest while the hand path was unaffected. This suggested to them the presence of compensatory joint rotations to minimize deflections in the hand trajectory, independent of whether the trunk was recruited or mechanically blocked. Thus this study supported the findings of Pigeon et al. (2000) that the involvement of the trunk is compensated by appropriate joint rotations. Adamovich et al. (2001) suggested that the integration of additional (in this case: trunk) degrees of freedom into the movement is based on afferent (proprioceptive and vestibular) signals. No central commands are issued for the compensatory arm movements. Instead the control system modulates the degree of compensation by “gating” the afferent signal elicited by the trunk motion. Through this control system, an appropriate contribution of trunk motion is provided to the hand transport. The involvement of afferent information provided by vestibular and proprioceptive pathways was investigated recently by Tunik et al. (2003) and Raptis et al. (2007). Tunik et al. (2003) investigated arm and trunk coordination in deafferented patients who had preserved vestibular afferent information. The authors found that patients were able to maintain the invariance of hand trajectory independent of trunk involvement. They explained the results by suggesting that patients had used
vestibular signals evoked by the head motion following the trunk flexion. Following the hypothesis presented in Tunik et al. (2003), Raptis et al. (2007) have investigated arm and trunk coordination in patients with vestibular deficits. Their results demonstrated that the majority of patients with vestibular deficits could not perform the arm angle rotations needed when the trunk was involved in the pointing tasks. The arm angle rotations are essential to compensate for the involvement of one more degree of freedom in the task (trunk displacement) in order to keep the trajectory invariant. Therefore, patients with vestibular deficits had their hand position and trajectory disturbed when the trunk was involved in the task, while healthy subjects where able to integrate the additional trunk movement without disturbing their hand trajectories. The authors concluded that vestibular afferent signals are essential for the motor system in order to produce and coordinate arm and trunk movements and keep hand trajectory properties invariant.

In healthy subjects, when the trunk is involved in reaching, its contribution to the endpoint movement occurs near the end of reach as the hand approaches the target (Rossi et al. 2002). Thus, the healthy nervous system uses a specific strategy to add the trunk movement when reaching to targets beyond a critical distance. It has been reported that the threshold for trunk recruitment during such reaching movements is lower in hemiparetic subjects (Levin et al. 2002). This study examined reach-to-grasp movements towards targets, located at four different distances, in 11 healthy and 11 hemiparetic subjects. Although healthy subjects did not use the trunk to reach closer targets (within arm’s length), hemiparetic subjects used considerable trunk motion, corresponding to the amount used by healthy subjects to reach farther targets (beyond arm’s length). They suggested that this increased trunk involvement might be due to the need to preserve trajectory smoothness or to limit movement errors, since the ability to extend the arm into extrapersonal space is limited in hemiparetic subjects. Tunik et al. (2004) have investigated the arm and hand response to trunk perturbations in patients with Parkinson’s disease. They observed that some patients with Parkinson’s disease were unable to keep their hand trajectories invariant when the
trunk movement was arrested. In addition, when patients tried to perform the appropriate angle rotations in the arm to compensate the trunk perturbation, those angle rotations had longer latencies than the ones seen in healthy control subjects. Based on their findings, the authors have suggested that the basal ganglia are important for organizing adaptive behaviour.

All the above studies support the idea that there exists a complex relationship between the arm and trunk movement during reaching movements. A disruption of this relationship can lead to abnormal recruitment of the trunk and/or an impaired UL function.

2.6. Insights into reaching movement performed by typical developing children and children with Cerebral Palsy.

2.6.1. Reaching in TD children:

TD infants learn to reach at about 4-5 months of age. At this time, their hand trajectories are jerky and chaotic, similar to spiral movements (Von Hofsten 1979, 1982, 1991; Fetters and Todd 1987; Thelen et al. 1996; Konczack and Dichigans 1997). Adult reaching patterns are considered well established when compared to infants, since adults maintain straight and smooth hand motion towards the target. According to Thelen et al. (1996) and van der Heide et al. (2005), reaching in young infants is still immature because their trajectories are still coupled with energetic and biomechanical constraints of movement execution. Thus they can not be adapted to environmental or task-specific constraints. In contrast, adults are able to maintain a smooth hand path independent of movement speed. They are also capable of varying hand motion with corresponding visual information in different postural contexts and task demands. The development of grasping and prehension has been well-studied through the analysis of manipulatory forces during a grip task and it has been found to gradually develop until the teenage years (Brooks 1986, von Hofsten and Ronnquist 1988, Gordon et al. 1994, Forssberg et al. 1991). However, it is not the
goal of this literature review to address the grasp component of reaching movements.

Von Hofsten (1979) analysed reach trajectories in 5 infants aged from 12 to 18 weeks old. He described that reaches could be divided into movement elements or movement units. The definition of a movement unit is based on a given velocity profile. Each unit consists of an acceleration and a subsequent deceleration phase. When a movement starts to accelerate again, a new movement unit is defined. Speed valleys demarcate the borderlines between units.

As was discussed in the previous section, adult reaching movements are characterized by a bell-shaped velocity profile, and thus by one movement unit. With age, children decrease the number of movement units during reaching (Von Hofsten 1979). In addition, as the number of movement units decrease, the first movement unit occupies a larger proportion of the reach, so that movement is formed by one acceleration and one deceleration. By 2 months of age following the onset of reaching, trajectories become more smooth and fluent, formed basically by one movement unit (Von Hofsten 1991; Thelen et al. 1993; Konczack 1995).

Thelen et al. (1996) suggested that movement units could be deliberate corrections to the trajectory at a higher level. Thus, the segmented trajectory could result from the infant’s inability to generate a virtual trajectory. Overall, studies suggest that in order to acquire better performance during reaching, infants must learn several levels of control, which consist firstly of a planning level and later, the stabilization of a programming, or execution, level. Although infants improve their performance in reaching within the first year of life, the development of coordination in reaching and the stabilization of reaching patterns that leads to stereotyped adult performance is not yet acquired (Konczak et al. 1995). Konczak and Dichgans (1997), with the goal of identifying when infants achieve adult-like consistency of kinematic performance in reaching, studied nine infants longitudinally from the onset of reaching (5 months) up to the age of three years and compared these results to those from four healthy adults. The task consisted of reaching for a stationary object placed at shoulder height. The study found that
straightness of trajectory increased over 90% by 3 years of age with respect to the initial value at the age of 5 months, but still differed from adults. Variability between trials decreased with age but at 3 years old was still higher than adults. Unimodal endpoint velocity (one movement unit) became predominant by two years of age. Temporal coordination between shoulder and elbow fluctuated largely during the first year. At 5 months of age, early reaches showed a pattern of peak velocity of elbow extension preceding shoulder motion. Adult reaching movement was characterized by a temporal pattern of shoulder flexion followed by elbow extension. Only by 24 months of age did infants demonstrate, in the majority of trials, an adult temporal sequencing between shoulder flexion and elbow extension. These authors concluded that endpoint trajectory smoothness emerges with the evolution of interjoint coordination. The underlying synergies that are the basis of invariant interjoint patterns are not established when infants start to reach. Nor are they acquired by the age of three years. Rather they had to be achieved during ontogenesis. This suggested that the developing nervous system employs synergies to reduce both the number of controlled movement parameters and the amount of afferent information necessary to generate and guide movement. The fact that the study found an emergent temporal coupling between limb segments is an indication of this constraint used by the CNS. However, the data cannot be conclusive on this issue since it only investigated reaches to a single target distance. In order to determine if this coordination is a general strategy of the CNS, reaches to different target locations should also be investigated.

Schneiberg et al. (2002) have investigated the development of coordination during reach-to-grasp tasks in TD children. The authors investigated development in 38 TD children aged from 4 to 11 years with a cross-sectional design, and compared them to 9 healthy adults. The reach task was performed in three different workspaces relating to arm length. The goals of the study were twofold: the first was to determine when children acquired mature patterns of reaching, obtained with movement quality analysis through kinematic variables. The second was to provide normative values for reaching through kinematic
variables in school-aged children for future comparisons with children with CP. The authors found that hand trajectories become smoother, straighter and less variable with age and children acquired an adult pattern of hand trajectories around 6-7 years for smoothness, and 8-9 years for straightness. The authors also reported that young children used more trunk displacement to reach targets placed within arm’s length than adults. In addition, a mature pattern of trunk displacement and intra and inter variability patterns of arm interjoint coordination were only acquired by the age of 10-11 years. The results of this study reaffirm the notion that development is not linear, but rather is composed of phases of increase or decrease in some motor parameters (Smith and Thelen 2003). The authors further suggest that the nervous system prioritizes hand trajectory, since it was the first variable to be developed and stereotyped when compared to interjoint coordination (elbow extension vs. shoulder-horizontal adduction), which remained highly variable at 11 years. In addition, they observed that trunk displacement decreases with age. Therefore, they have suggested that trunk involvement was excessive in young children when compared to the adult group because the children had not yet acquired the maturation in cortical areas required to integrate for arm and trunk synergy.

By increasing knowledge regarding the development of movement quality in reaching and grasping activities in TD children, it will be possible to characterize the same development in children with CP with more quantitative and objective measurements. The characterization of movement quality in reaching tasks through kinematic analysis provides researchers and clinicians with a detailed description of movement performance. Therefore, the use of motor quality analysis should be incorporated into the description and evaluation of abnormal motor development. In addition, the use of motor quality analysis as a measurement in children with CP might increase the sensitivity to detect changes in UL function due to rehabilitation interventions.
2.6.2. Movement quality of reaching movements in children with CP.

The control of hand path during reach-to-grasp activity is possible due to the interaction of multiple systems and cortical sensorimotor associations (Jeannerod 1990). Movement quality is concerned with movement performance or how well an activity is performed taking into account normative data from typical populations (Van de Winckel et al. 2006). The assessment of movement quality in UL activities refers to the measurement of range of motion, hand trajectories, interjoint and intersegment coordination, muscle contraction patterns, and postural adjustments. Many studies have investigated the movement quality of reaching in children with CP. Overall they have shown that reaching in children with CP is characterized by increased movement duration (MacKey 2006), a decrease in range of motion of UL angles (Van Thiel and Steenbergen 2001), a lack of smoothness, or segmented hand trajectories (van der Heide 2005, Chang 2005), an increased variability in hand trajectories (Sanger 2006), a lack of anticipatory aperture of fingers when grasping (Ronnqvist and Rosblad 2007), and excessive trunk displacement (van Roon 2001, van Thiel 2001, Mackey 2006, Kreulen 2007).

The presence of segmented hand trajectories in children with CP has been reported in most studies investigating reaching movements in this population. Hand trajectories, as discussed in the previous section, might be planned by coupling visual and kinesthetic information. Recently, two studies have investigated, eye and hand coordination during reaching tasks in children with CP (Saavedra et al. 2008 and Verrel et al. 2008). Verrel et al. (2008) have reported that children with hemiplegic CP have eye movements as fast and accurate as their TD peers. However, when they performed the reaching tasks with their most affected hand they tended to increase visual monitoring of the hand movements through all phases of the task. The authors have suggested that this adaptation in hand-eye coordination was a form of compensation for sensory motor deficits. They have also reported that the children with CP spent longer times with their eyes on the target, which they had interpreted as increased attentiveness. Saavedra
et al. (2008) as well as Verrel et al. (2008) did not find differences in anticipatory gaze control between children with CP and TD children. However, Saavedra et al. (2008) have suggested that children with CP have problems adjusting eye movement and hand movement, which is a different interpretation than the one given by Verrel et al. (2008). They have suggested that children with CP can not voluntarily control undesired saccadic movements. Moreover, the authors had tried to investigate the movement of the hand without eye movement but it was impossible because children with CP kept moving their eyes towards the moving hand. Interestingly, the authors have reported that this finding was similar in TD children aged 4-6 years, and might suggest a delay in development in eye-hand coordination in children with CP.

Limited range of motion and excessive trunk displacement were also a consistent finding in the studies investigating reaching in children with CP. Van Roon et al. (2005) have demonstrated that when children with CP are asked to reach within their range of motion limits, they did not have excessive trunk displacement. Kreulen et al. (2007) investigated the relation of forearm movement rotations and trunk movement in teenagers and adults with CP. They have observed that during a maximal supination task (drinking task), subjects with CP were unable to perform the task without recruiting excessive trunk movement. The authors suggested that there is a relationship between forearm impairments detected during dynamic activity and compensatory trunk movement. Some authors have suggested that excessive trunk displacement in children with CP might be related to the task accuracy or the speed of arm movement (van Roon et al. 2004, van Roon et al. 2005). Other authors have suggested that children with CP recruit more trunk displacement than their TD peers because they showed both a delay in postural anticipatory reactions and abnormal recruitment of postural muscles during reaching (van der Heide 2004, Hadders-Algra 1999).

Most studies to date have provided a description of movement quality in different reaching tasks in children and adults with CP. Current experimental studies in UL movements in children with CP are available, including those with movement perturbation and different environmental aspects. These have focused
more on understanding the relation between different postural supports, sitting positions and the postural adjustments during reaching (van der Heide et al. 2004; Stavness 2006) or interlimb coupling in bimanual activities (Utley et al. 2004; Utley and Steenbergen 2006). More experimental studies, such as Verrel et al. (2008) and Saavedra et al. (2008), where high level of hand movement control is investigated are needed in order to understand the underlying mechanism behind sensorimotor disorders and the lack of movement quality in reaching in CP. No study to date has investigated the effect of rehabilitation intervention on upper limb movement quality. The assessment of movement quality is essential to avoid misinterpretations of motor compensations as improvement. Motor compensations are defined as atypical movement patterns. They are involuntarily elicited by the individual in order to compensate for a specific deficit. Therefore, motor compensations can be considered maladaptive movement patterns that are used by disable individuals to successfully accomplish an activity.

2.7. Rehabilitation strategies to improve UL function in children with CP: what is the evidence?

Currently, evidence-based practice is a subject of a considerable debate in all health areas. Evidence-based practice (EBP) or evidence-based medicine (EBM) was originated in Canada, at MacMaster University in Hamilton, Ontario by the pioneering work of David Sackett and colleagues in 1970 (Law and MacDermid 2008). Sackett’s (1998) definition of EBM is: “Evidence-based medicine is the conscientious, explicit, and judicious use of the current best evidence in making decisions about the care of individual patients. The practice of evidence-based medicine means integrating individual clinical expertise with the best available external clinical evidence from systematic research.” Although the ultimate goal of generating scientific evidence for treatments is the knowledge transfer to best practice, in reality the implementation of research findings in clinical settings is not an easy task (Saleh et al. 2008).
Most of the systematic reviews that have investigated the effectiveness of rehabilitation therapies to improve UL functions in children with CP are inconclusive regarding the existing level of evidence (Wasiak et al. 2004; Lannin et al. 2006; Hoare et al. 2007; Lannin et al. 2007; Anttila et al. 2008). However, due to financial and temporal constraints, it is becoming increasingly difficult to deliver a physical and occupational therapy intervention that does not have scientific support. According to Siebes et al. (2002), several factors have contributed to the lack of efficiency and effectiveness of existing therapeutic motor intervention programs for children with CP. Two of the most important factors are poor methodology quality, and deficiency in design, as well as a lack of instruments sensitive enough to detect small changes in movement quality or performance.

The criticisms about the methodological quality in the trials are generally related to the internal validity of findings, such as a lack of randomization or lack of blinding of the evaluator to the main outcome, as well as small sample sizes (Lannin et al. 2007; Anttila et al. 2008). Although randomized clinical trials (RCTs) are considered the ‘gold standard’ to prove treatment effectiveness, according to Law and MacDermid (2008) any such importance in the responsibility of RCTs to prove high-level scientific evidence has diminished the fact that some research questions are better fitted with other types of experimental designs. The authors have suggested 5 different levels of evidence, where the highest level 1, accounts for all aspects necessary to an optimal internal validity. As some aspects of internal validity are lost (e.g. randomization) the level of confidence that the evidence is valid decreases. Level 1 includes systematic reviews and RCTs, while the lowest level of evidence (level 5) is where there is no observation of patients. Level 5 is still commonly seen in clinical practice in rehabilitation. In this level, evidence is gathered based on an expert’s opinion without further research. While the authors agreed that the expert’s opinion is important to generate clinical questions, they reaffirm that it is only through hypothesis testing that high levels of confidence in treatment evidence can be obtained. Furthermore, it has been suggested that perhaps RCTs are not the
optimal design to determine the effect of interventions in children with CP due to the high heterogeneity of the group (Siebes et al. 2002).

Another explanation for the lack of scientific evidence in rehabilitation strategies to improve UL function is the type of outcomes used (Wasiak et al. 2004; Hoare et al. 2007). Most of outcomes are not sensitive to change or they are not appropriately designed to answer the study question. Undoubtedly, the choice of outcome is essential to detect efficacy of an intervention (Majnemer and Mazer 2004). Rather than simply having the required psychometric properties, an outcome used in children with CP should account for different aspects of function related to age during motor development, be associated with the type of question in the study (discriminative, predictive, or evaluative) and be used according to the dimensions proposed by the ICF (Majnemer and Mazer 2004).

To date, most of the studies that have investigated the efficacy of upper limb interventions in children with CP have focused on the treatment of motor impairments such as spasticity, reduced range of motion, weakness, sensory deficits and learned nonuse issues (Fetters and Kluzik 1996; Maenpaa et al. 2004; Charles et al. 2006; Deluca et al. 2006; Ozer et al. 2006; Russo et al. 2007; Wallen et al. 2007), which has been proved to be not strongly associated with changes in activity level (Damiano 2008). None of these clinical trials investigated the use of a task-oriented approach to treat UL function in children with CP. However, Charles and Gordon (2006) have proposed a Hand and Arm Bilateral Training (HABIT) where the training consists of structured practice using motor learning principles, as suggested by the task-oriented approach. They investigated the efficacy of HABIT training compared to a no-treatment or delayed treatment with RCT design in twenty children aged from 3½ years to 15½ years. The results were favourable to the HABIT treatment as detected by the Assisting Hand Assessment (AHA), frequency of hand use detected with an accelerometer, and the bimanual items of the Bruininks-Oseretsky test of motor proficiency. The limitations of the study were the difference in baseline between the two groups: the intervention group was more impaired. As well, the effect of
HABIT training in the control group, which was supposed to receive the intervention later, was not reported. However, based on the changes seen in the intervention group the authors have suggested that HABIT training should be further investigated with larger samples or different designs. Recently, the CanChild centre for childhood disability research has proposed a multi-centred randomized clinical trial to investigate function in children with CP by comparing a task context-focused approach to a child-focused approach. The trial is promising because the proposed intervention and outcomes will integrate the updated dimensions of the ICF (Law et al. 2007).

In conclusion, to date there is no strong evidence (1a – Systematic review of RCTs or Meta Analysis) of physical or occupational therapy treatments for improving UL function or activity in children with Cerebral Palsy. However, the most promising rehabilitation strategies are serial castings, constraint induced therapy, HABIT, and task oriented treatments. Therefore, more randomized clinical trials with new strategies of treatment, or better methodological designs, are required.

2.8. Constraint Induced Therapy & movement quality

Physical constraint of the less-affected upper limb as a rehabilitation intervention is a promising strategy for improving hand and arm function in children with hemiplegic CP (Boyd et al. 2001). There are three types of constraint therapy: constraint induced therapy (CIT), modified constraint induced therapy (mCIT) and forced used therapy (FUT) (Hoare et al. 2007). In all types, movements of the less-affected arm are restricted and practice with the more-affected arm is encouraged. While CIT protocols include more than three hours of therapy per day for at least two consecutive weeks, in mCIT protocols, the amount of therapy is usually reduced to less than three hours a day (Hoare et al. 2007). CIT includes adaptive task practice or ‘shaping’ which may or may not be incorporated in mCIT. In FUT approaches, aside from the constraint protocol, no additional therapy is provided for the affected arm. While studies have provided
evidence of increased use of the affected arm after CIT, mCIT and FUT, it is unknown whether this is accompanied by improvements in movement quality at the kinematic level (e.g. increased range of active movement, greater movement smoothness and better interjoint coordination) or in activity level (e.g. self-care). Improvements in movement quality using constraint therapy would imply that the movement patterns used to perform tasks become more like those seen in TD children.

One goal of constraint-induced therapies is to prevent learned nonuse of the more affected upper limb (Taub et al. 2006). Following hemispheric brain lesions, anatomical and neurophysiologic changes occur in the intact hemisphere paralleling the changed function of the contralateral body side as it compensates for the loss of the function of the hemiparetic limbs (Allred and Jones 2008). These findings suggest that the intact hemisphere might prevent improvement of function in the impaired body side (Allred and Jones 2008). In addition, during the period of improvement from unilateral brain lesions, in adult human and animal, studies have shown that new atypical movement patterns emerge during reaching tasks attempted with the hemiparetic arm (Levin et al. 2002; Alaverdashvili et al. 2008). These new motor patterns are compensatory and have recently been termed ‘learned bad use’ (Alaverdashvili et al. 2008). The emergence of atypical patterns is thought to hamper improvement since they may be reinforced with repetitive practice instead of allowing more typical movement patterns to appear (Allred & Jones, 2008; Alaverdashvili et al. 2008).

As discussed before in section 2.6.2 children with CP use alternative movement patterns during reaching and grasping compared to TD children. The effects of CIT, mCIT and FUT on movement quality are difficult to evaluate since most studies describe improvements in terms of functional task accomplishment (i.e., task completion time, number of tasks completed) without consideration of the quality of task performance (Charles and Gordon 2005). Hoare et al. (2007) found a trend for a positive treatment effect using modified constraint induced therapy (mCIT) detected by the QUEST, in a single non-randomized trial a strong positive effect was found for the mCIT detected by the AHA. Charles and Gordon
(2005) reviewed 15 trials that used CIT or a modified version, none of the trials was classified as RCT with large sample size or level 1b of evidence (Law and MacDermid 2008).

2.9. Task oriented: a proposed model of intervention for PTs and OTs to treat UL deficits in children with CP based on the ICF levels.

As discussed in a previous section, no study to date applied task oriented intervention to improve UL movements in children with CP. Task oriented intervention is based in biomechanical analysis of movement and in motor learning theories (Shumway-Cook and Woollacott 2001). The goal of task oriented is to treat impairments during functional activities. Task oriented intervention comprises repeated practice, and goal oriented movements that are selected according to individual deficits. The therapist, when using task oriented intervention, makes use of biomechanical information to develop environment constraints during the training. In order to plan the task oriented approach used as treatment in this thesis, a model of treating disability and function was adopted based on the international classification of functioning, disability, and health (ICF, WHO 2001; Grimby and Smedby 2001). The ICF is a framework proposed by the World Health Organization for describing health and health-related states, outcomes and determinants. The ICF model has two parts: Part 1: Functioning and Disability, which is comprised of three components of health: body function & structures, activities, and participation; and Part 2: Contextual factors which are formed by environmental and personal factors (Shumway-Cook and Woollacott 2007; Palisano et al. 2006). If the UL deficits in CP are put in the context of the ICF, the body function level will include the following impairments: spasticity, weakness, muscle stiffness, poor postural control, decreased range of motion, and sensory deficits. In the activity limitation level it will include problems in reaching, grasping, feeding, writing and other self-care tasks. On the participation level, a child with CP will have social restrictions in playing, learning, and probably in practicing some sports. The environmental factors can comprise the
school, facilities at school and home, winter season (putting on winter clothes), and opportunities to practice. The personal factors are the child’s attention, motivation, engagement, attitude towards therapy, and caregiver assistance. When using the ICF framework in a clinical decision, the most important factor is to understand the degree of interaction among the different levels. One should understand the association of contextual factors and participation, disability and functional limitation, as well as functional limitation and impairment. Task oriented intervention is consistent with the ICF model because it treats function in relation to disability and motor impairment (Shumway-Cook and Woollacott 2007).

A task oriented intervention with the goal of improving UL deficits in children with CP should focus on treating or minimizing impairments in order to maximize function and reduce disability. By developing efficient task-specific strategies and adapting goal-oriented movement to different tasks and environments, the therapist will provide a variety of conditions to practice UL function that will be task dependent and related to the main underlying impairments which may be the cause of disability. The first step is to assess the child’s performance and capacity in tasks that involve UL movement. The second step will be a detailed examination of the strategy used by the child while performing the movement. The third step is to determine the association in the multilevel (impairment, activity, environment, personal factors) with the disability in order to design a specific treatment. The task oriented approach is a promising intervention because each treatment design is specific for an individual. In other words, each child will receive a treatment focusing on their own needs. It is important to remark that a task oriented approach differs from a task-specific training approach (Shepherd 1995, Shumway-Cook and Woollacott 2007). In the task oriented intervention, a single session includes treatment focusing on impairment level, strategy or key movement components level, and the practice of the task or functional level. In contrast, the task specific training focuses on practicing the task itself and its specific component(s), with little to no focus on the underlying impairments or the strategy of movement.
Several studies have demonstrated that reach and grasp movement components are determined by the goal and context of the activity, and that the relationship between underlying impairments and arm and hand functions may be task dependent (Wu et al. 1994; Gordon and Duff 1999). In addition, it is well-documented that in order to learn or relearn a motor skill, the amount and type of practice plays a determinant role in task performance (Schmidt et al. 1989; van Dijk et al. 2005; Cirstea and Levin 2007). Thus, an intervention for UL using a task oriented approach, where impairments are minimized, key components of reach-to-grasp movement are trained, and application to function is required, may be a promising method to overcome the lack of evidence in the effectiveness of treatment for UL function in children with CP.

2.10. Trunk restraint strategy

The use of a trunk restraint (TR) strategy during upper extremity training with a task oriented approach is justified on the basis of findings from previous studies where children and adolescents with CP performed reaching and grasping activities and their UL movements occurred together with excessive trunk movements (Steenbergen et al. 2000; Volman et al. 2002; van Roon et al. 2004; van Roo et al. 2005; Mackey et al. 2006; Coluccini et al. 2007; Kreulen et al. 2007). The excessive trunk movement observed in children with CP in those studies is defined as abnormal because it is not observed in age-matched control groups. Moreover, the amount of trunk movement observed in children with CP can be twice the amount of trunk movement observed in normally developing children (van Roon et al. 2004, 2005, Mackey et al. 2006).

The idea to integrate a TR strategy during UL training in children with CP came from previous studies in adults with hemiparesis that reported benefits of trunk restraint training (Levin et al. 2002; Michaelsen and Levin 2004; Michaelsen et al. 2006). It is supported by the subsequent positive results in arm and hand motor performance when the child’s trunk is passively restrained by the therapist (clinical reports) or by an external device (Butler 1998; Stavness 2006),
or when the reach performance improved when the trunk is fixed (van Roon et al. 2005). Therefore, the improvements in arm and hand functions when the trunk is restrained are findings that support the idea that excessive trunk displacement during upper extremity activities might be undesirable and harmful to optimal performance.

Stavness (2006), in a systematic review, found support evidence that in order to improve UL skills in children with CP, they should be in a functional sitting position, which should be individually established. The importance of arm and trunk coordination to transport the hand was reported in many studies in the adult population (Adamovich et al. 2001; Ghafouri et al. 2002; Rossi et al. 2002; Tunik et al. 2003; Raptis et al. 2007).

By restraining the excessive movements of the trunk during reaching and grasping training, more pertinent somatosensory input from the arm joints can be provided and used to modulate the reaching pattern. During task oriented activities, children who have their trunk restrained will be unable to use excessive trunk movement to help bring the hand to the object. They will then have to use the appropriate UL joint rotations in order to accomplish the task. Therefore, the trunk restraint might expose the child to appropriate task related somatosensory information that will lead to an improvement in UL skills and function (Hadders-Algra 2001; Shumway-Cook and Woollacott 2007).
2.11. Summary

Children with CP are extremely heterogeneous in terms of etiology and clinical features. The diversity of symptoms among CP syndromes is a challenge for different areas of health research in terms of definition and classification. Despite the efforts of many studies in examining rehabilitation strategies to improve UL function in children with CP (Boyd et al. 2001; Antilla et al. 2008), the confidence in the validity of these studies’ evidence is still moderate to low (Charles and Gordon 2005). One of limitations here is the lack of strong evidence related to the type of outcome. Most of the studies’ outcomes are not sensitive enough to detect change (lack of responsiveness), or they are not related to age. Nor did they describe the movement quality or how the activity is performed. The description of movement quality is important, because early brain injuries are more susceptible to ‘maladaptive’ plasticity, which leads to abnormal movement behaviors. Motor compensations, such as excessive trunk displacement, have been described to occur during UL movements in children with CP. A better method for examining treatment efficacy in children with CP might be kinematic analysis. Kinematic analysis has been used to describe the acquisition of reaching and grasping activities during different stages of motor development in TD children. Therefore, by analyzing how movement is performed, improvement can be more fairly assessed compared to more subjective measures of task accomplishment. The assessment of movement quality is essential to avoid misinterpretations of motor compensations as improvement. Overall, one of the more promising rehabilitation interventions to improve UL movements currently available is CIT. However, most of the studies investigating the effect of CIT did not investigate movement quality using kinematic analysis. Finally, the updated concepts of the ICF and the demand to use outcomes and treatments that follow this model, necessitate the exploration of task-oriented intervention, which remains an untested means of improvement of UL function in children with CP.
2.12. Rationale

The lack of available evidence in the literature favoring the effect of rehabilitation interventions in UL function as well as the increasing demands that professionals apply evidence-based practice in clinical settings, are factors suggesting that there is a need to develop more studies with different designs and innovative treatment interventions in order to improve UL function in children with CP. Deficits in UL function are a common problem in children with CP, and is often accompanied by limitations in activity and participation. There are three main problems that contribute to the lack of progress in providing evidence in rehabilitation strategies to improve UL function in children with CP: 1. poor methodological quality and deficiency in design in most of the existing clinical trials; 2. lack of instruments sensitive enough to detect small changes in motor ability or able to describe movement quality; 3. interventions and strategies not addressing the dimension levels of the ICF.

Due to the broad heterogeneity inherent in CP, and the diverse factors that can influence UL activities, it has been suggested that grouping this population is not the optimal research design for investigating treatment effects (Siebes et al. 2002). The majority of available RCTs that aimed to investigate the efficacy of UL interventions on this population did not report or lacked the power to prove treatment effectiveness (Hoare et al. 2007). In addition, it has been questioned if generalization of treatment effects by group comparisons, especially in children with CP, is transferable to clinical practice (Saleh et al. 2008; Bartlett 2008). Single subject research design, with a highly controlled internal validity, might be an optimal approach for investigating treatment effects in children with CP.

The lack of outcomes which are able to detect changes (those that are highly responsive) is a major problem in research and clinical practice involving children with CP. In addition, outcomes should be employed according to the dimension level investigated (e.g. function - range of motion: goniometer; activity – hand skills: AHA). Although, pediatric outcomes should account for development issues, little is known about the flow of development in children
with CP. Some studies have demonstrated that development depends on the level of severity of CP: some children can become worse in their movements as they grow. Furthermore, the optimal outcome measurement should describe movement quality or how the task is performed, in order to avoid the misinterpretation of motor compensations as improvement. Motor compensations are defined as atypical movement patterns. They are involuntary elicited by the individual in order to compensate for a specific deficit. Therefore, motor compensations can be considered maladaptive movement patterns that are used to successfully accomplish an activity. In addition to the outcomes problem, some interventions that apply the interaction of some levels of the ICF model, such as the task oriented approach, remain uninvestigated. Ultimately, strategies based on motor learning approaches and neuroplasticity theories should be tested in conjunction with kinematic analysis to verify if they are able to improve UL movement quality in children with CP.

The research projects in this thesis are designed to determine the effect of two promising rehabilitation strategies for UL movement quality: 1. the use of constraint in the less affected arm together with conventional occupational therapy; 2. the use of a trunk restraint strategy together with task oriented intervention. The main outcomes used to measure movement quality are kinematic variables of a functional reaching task. The kinematic analysis of reaching has been investigated in TD children in previous studies, providing normative data for further comparison. In addition, a test-retest analysis was conducted on the main kinematic variables, in order to determine the ones that had the highest reliability in order to better discriminate clinical changes.

2.13. Objectives

The primary objective of this prospective study was to determine the effect of two rehabilitation strategies in UL movement quality: arm constraint and trunk restraint. The UL movement quality was measured by kinematic analysis of
a functional reaching task: a self-feeding simulation. Only the reaching phase of the task was analysed.

2.13.1. Specific objectives

1. To characterize movement quality measured with kinematic analysis before and after mCIT.
2. To determine if changes in clinical assessment following mCIT are accompanied by improvements in kinematic patterns.
3. To determine the most reliable kinematic variables for use in clinical trials, especially those best able to define individual differences and changes after treatment.
4. To determine whether UL movement quality may be better improved by task-oriented intervention alone or when coupled with a trunk restraint strategy.

2.13.2. Hypotheses

In line with the objectives (2, 3, and 4) the following hypotheses were tested:

1) mCIT therapy will result in improvements in movement quality detected by clinical and kinematic outcomes.
2) Kinematic variables will have high test-retest reliability.
3) A better effect on movement quality with a decrease in movement compensations will be detected in children who received task-oriented intervention with trunk restraint strategy than in children who received task-oriented intervention alone.
4) A Task-oriented intervention will improve UL movement quality in children with CP, and that these improvements would be maintained at three months following the end of the intervention.
CHAPTER 3

INTRODUCTION

The primary objective of this thesis is to determine the effect of two rehabilitation strategies in UL movement quality: arm constraint and trunk restraint. This first manuscript, therefore, investigated the effect of arm constraint in a context of mCIT in three young children with mild hemiplegic CP. The sample used in this manuscript was a sample of convenience by the Institutional Review Boards of Ste Justine Hospital to verify the feasibility of mCIT in clinical practice. Prior to application of the mCIT in the clinical setting, the safety of the method and the tolerance of young children with CP to the intervention needed to be investigated.

The following objectives were addressed in this paper: To characterize movement quality measured with kinematic analysis before and after mCIT, and to determine if changes in clinical motor impairment following mCIT were accompanied by improvements in kinematic patterns. The outcome measurements used were kinematic variables collected during a functional reaching task, self-feeding simulation to three different distances according to the child’s arm length. In addition the movement quality was assessed by the QUEST scale. Children were assessed two times before the arm constraint was applied. A second baseline assessment was made after the child had received two weeks of intensive standard occupational therapy without any arm constraint. The second baseline assessment served to test if differences in movement quality could occur because of intensive therapy alone. Following this second baseline assessment, an additional three weeks of intensive therapy using mCIT was given. At the end of this period, a post-intervention assessment was made to test the effect of mCIT. One month after treatment the child was again assessed to test carry over effects.
EFFECT OF MODIFIED CONSTRAINT-INDUCED THERAPY ON KINEMATIC MEASURES OF REACH TO GRASP MOVEMENT IN CHILDREN WITH CEREBRAL PALSY – THREE CASE REPORTS

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3.1. Abstract

Therapies to improve arm motor function using constraint of the less-affected arm in children with hemiparesis have sparked interest in rehabilitation researchers and clinicians. While constraint therapies may improve functional ability in the affected limb, few studies have assessed changes in movement quality using kinematic analysis. We characterized arm and trunk movement during reaching in three children (2-5yrs) with mild hemiparesis due to cerebral palsy before and after 3 weeks of modified constraint induced therapy using a repeated measures case (AABA) design. Main outcome measures were quality indicators of trajectory, joint range and trunk displacement and an arm impairment measure (QUEST). All children improved at least one quality outcome and two children improved QUEST. However, all children also increased compensatory trunk displacement. Although clinical improvements (QUEST) may be accompanied by some improvements in arm motor patterns during reaching, the possibility that compensatory movements may accompany these improvements should be considered.

Key words: Cerebral Palsy, rehabilitation, upper extremity, reaching, kinematics, forced use, compensation
3.2. Introduction

Physical constraint of the less-affected upper limb as a rehabilitation intervention is a promising strategy for improving hand and arm function in children with hemiplegic cerebral palsy (Eliasson et al. 2005; Charles et al. 2006; Deluca et al. 2006; Hoare et al. 2007). There are three types of constraint therapy: constraint induced therapy (CIT), modified constraint induced therapy (mCIT) and forced used therapy (FUT). In all types, movements of the less-affected arm are restricted and practice with the more-affected arm is encouraged. While CIT protocols include more than three hours of therapy per day for at least two consecutive weeks in mCIT protocols, the amount of therapy is usually reduced to less than three hours a day (Allred and Jones 2008). CIT includes adaptive task practice or ‘shaping’ which may or may not be incorporated in mCIT. In FUT approaches, aside from the constraint protocol, no additional therapy is provided for the affected arm. While studies have provided evidence of increased use of the affected arm after CIT, mCIT and FUT, it is unknown whether this is accompanied by improvements in movement quality at the kinematic level (i.e., increased range of active movement, greater movement smoothness and better interjoint coordination). Improvements in movement quality would imply that the movement patterns used to perform the task become more like those seen in typically-developing (TD) children. There is much debate about the extent to which motor gains result from improvements in movement quality and/or the development of alternative or compensatory movements and how rehabilitation influences these processes. Part of the problem is the lack of consensus among clinicians and researchers of different disciplines on the definitions of improvement of movement quality and compensation (but see Levin et al. 2008, subm). Although most fundamental researchers studying neuronal plasticity and brain reorganization agree on the definition of improvement/compensation at the neuronal level, this distinction has not been well-defined at the motor performance and functional levels. This has led to some confusion in the interpretation of the efficacy of different treatment interventions; often leading to equivocal results in which changes at each level are mutually confounded.
Kinematic measures provide the means by which changes in movement quality can be distinguished from the presence of motor compensation. Detailed kinematic movement analysis helps to interpret the results of clinical performance better than functional scales that do not distinguish between improvement and compensation of motor patterns. One goal of constraint-induced therapies is to prevent learned nonuse of the more affected upper limb (Taub et al 2006). Following hemispheric brain lesions, anatomical and neurophysiologic changes occur in the intact hemisphere paralleling the changed function of the contralateral body side as it compensates for the loss of the function of the hemiparetic limbs. These findings suggested that the intact hemisphere might prevent improvement of function in the impaired body side (Allred and Jones, 2008). In addition, during the period of improvement from unilateral brain lesions, animal and human studies have shown that new atypical movement patterns emerge during reaching tasks attempted with the hemiparetic arm (Levin et al. 2002; Alaverdashvili et al. 2008). These new motor patterns are compensatory and have recently been termed ‘learned bad use’ (Alaverdashvili et al. 2008). The emergence of atypical patterns is thought to hamper improvement since they may be reinforced with repetitive practice instead of allowing more typical movement patterns to appear (Allred and Jones 2008; Alaverdashvili et al. 2008).

Children with spastic forms of CP used alternative movement patterns during reaching and grasping compared to TD children. Most studies using kinematic or kinetic analysis have focussed on children older than 8 and report a lack of movement smoothness (more movement units in the trajectory), a decreased active range of joint movement, a disturbed elbow-shoulder interjoint coordination (segmentation), incoordination between reaching and grasping components, inflexibility in selection of hand orientation for grasping and the use of excessive trunk movement during reaching and grasping. In younger children with CP, deficits in trajectory formation and movement smoothness have been described (Levin and Jobin 1998). Children with CP may also use more trunk
displacement during reaching compared to age-matched TD children (Schneiberg et al. 2004).

The effects of CIT, mCIT and FUT on movement quality are difficult to evaluate since in most studies describe improvements in terms of functional task accomplishment (i.e., task completion time, number of tasks completed) without consideration of the quality of task performance. We addressed this question by documenting if improvements in clinical motor impairment following mCIT were accompanied by improvements in kinematic patterns in young children with mild CP. Preliminary data have appeared in abstract form (Schneiberg et al. 2004).

3.3. Methods

Participants

Ethics approval was obtained to recruit a convenience sample of three children for this study. Children could participate in the study if they had CP affecting one arm more than the other and had a GMFCS Level of I. Families/children signed informed consent forms approved by the Sainte-Justine Hospital Ethics Committee.

Child 1 (Jason) 

Jason is a boy, aged 2 ys 6 mos at the time of entry into the trial. He was born from a normal pregnancy. At 2.2 mos, his parents noticed that he used his left hand less than his right and by 10 mos of age, a diagnosis of left-sided hemiparesis due to antenatal stroke was confirmed with CT scan. He had moderate spasticity in the left arm which tended to adopt a flexor synergy with closed hand and adducted thumb at rest or on exertion. Active elbow and shoulder joint ranges were mildly reduced as was active finger flexion range with the wrist extended. Functionally, he could eat independently using his fingers or a spoon placed in the right hand and could drink from a glass. He could use his left hand to manipulate objects if required but movements were not coordinated and he had difficulty grasping and releasing objects.

1 Children’s names are fictional to protect their identity.
**Child 2 (Linda)**

Linda is a 3 yr 4 mo old girl, born from a normal pregnancy. A CT scan done at 12 mos showed a left hemispheric atrophy with parietal encephalomalacia suggesting a left middle cerebral artery stroke. She had mild spasticity in her right arm but lacked dexterity and coordination during activities requiring fine motor tasks of the right hand. For example, in a timed task she could stack 8 blocks with the left hand but only 2 with the right hand. She had difficulty dissociating her fingers and grasping and releasing small objects (e.g., ¼” pegs).

**Child 3 (Beth)**

Beth is a girl aged 4 yrs and 4 mos at the time of the study, also born of a normal pregnancy. When Beth was 2 months old, greater use of the right than the left arm was noticed. MRI revealed ischemic lesions in the right internal capsule the parietal subcortical white matter. Prior to the trial, Beth used her left arm spontaneously but had incomplete forearm supination. She had mild spasticity and tended to adduct her left thumb leading to prescription of a thumb abduction orthosis. The left arm was well-integrated during play activities although she tended to mildly neglect this arm during activities of daily living. She could perform precision grasping but had difficulty dissociating and adjusting her fingers during manipulative tasks. She could screw and unscrew the lids of jars but had difficulty with scissors and stringing beads.

**Study design**

We used a single-case, repeated measures (AABA) design in which post-test and one-month follow-up data were compared to data from 2 pre-tests for each child (Fig. 1A). In each pre-test session, arm and hand impairment were evaluated with a clinical scale and arm and hand kinematics during seated reaching tasks were recorded. Following the first pre-test evaluation, each child had three 45 min sessions of occupational therapy (OT) per wk for 5 wks focusing on arm activities. After 2 wks of therapy, a second pre-test evaluation was done. Then,
the less-affected arm was constrained with a detachable progressive bi-valve cast encompassing the forearm, wrist and hand (Fig. 1B). The cast was worn continuously for the remaining 3 wks during which time the same therapy schedule of three 45 min sessions of OT per wk was continued. The cast was removed for hygiene and sleeping. As a precaution against injury from falling, each child wore a helmet when casted. A post-test evaluation was done at the end of the 5-wk period and repeated one month later (follow-up) to evaluate carry-over effects.

Assessments

Pre- and post-test evaluations were administered by a therapist not involved in delivering the treatment intervention. The valid and reliable Quality of Upper Extremity Skills Test (QUEST) was used as a clinical measure of arm and hand impairment (DeMatteo et al. 1992). QUEST domains are dissociated joint movements, grasping, weight bearing through the arm and protective extension. Test scores range from a negative value if all items are atypical to a maximum score of 100.

For the kinematic measures, the child reached and grasped a 2 cm³ block (target) in a simulated self-feeding task. Only the reaching movement was analysed, in order to minimize the numbers of markers placed on the child’s arm. For standardization, targets were placed in child’s body midline at distances proportional to the child’s arm length (2/3, 1 and 1+2/3 arm’s length, Fig. 1C). These distances were chosen since reaching to each target is accomplished by different patterns of arm and trunk movement (Schneiberg et al 2002). For example, in TD children, reaches to T1 and T2 require very little trunk movement, while reaches to T3 require trunk movement to assist arm displacement. A block of 10 trials was recorded for each target and blocks were performed in a random order. Children reached from a sitting position with their back unsupported. The table height was adjusted to the height of the child’s elbow. The position used for the first pre-test was reproduced for each child in each of the 4 evaluation sessions.
Kinematic data acquisition and analysis

Movements were recorded (5s, 100Hz) using a three dimensional optical tracking system (Optotrak, Northern Digital, Model 3010, 0.01 mm measurement error) with 8 infra-red emitting diodes (IREDs). IREDs were placed on the distal part of the second phalange of the index finger (defined as the movement endpoint), tip of the thumb, hand (middle of the second metacarpal), wrist (ulnar styloid), elbow (lateral epicondyle), shoulders (acromions) and trunk (sternum). Positional data from the IREDs (x,y,z) were low-pass filtered with a 5th order Butterworth (10Hz cutoff) filter. This is a standard frequency for filtering kinematic data since physiological movement is normally made between 6-10 Hz. Endpoint and trunk tangential velocities were computed from the velocity vector magnitude, obtained by differentiation of the positional data (5-point central difference numerical differentiation). To diminish measurement variability, markers were placed on well-defined bony landmarks that were standardized within each child.

Because of the limited marker set, we used vector analysis instead of rigid body calculations to compute joint angles. This marker configuration was chosen to avoid encumbering the children during data collection. Thus, joint rotation calculations of elbow flexion/extension and shoulder horizontal adduction/abduction did not include the influence of rotational movement. Likewise, trunk displacement measurement was restricted to the transversal (x,y) and sagittal (y,z) directions.

Kinematic parameters measured were: 1) Trajectory smoothness, defined as the number of peaks in the tangential velocity trace. A peak was defined as a maximum velocity proceeded/followed by increasing/decreasing values for at least 20 ms. 2) Trajectory straightness, measured as index of curvature (IC), defined as the ratio of the actual endpoint path length to that of an ideal straight line. Using this measure, a straight line has an index of 1 and a semi-circle has an index of 1.57. 3) Ranges of elbow flexion/extension and shoulder horizontal adduction/abduction angular motion. Elbow angle was computed as the dot
product of the vectors defined by coordinates of appropriate markers placed on
the wrist, elbow and ipsilateral shoulder. Shoulder horizontal adduction/abduction
angle was measured as the horizontal projection of the angle between vectors
defined by the ipsilateral and contralateral shoulder markers and the ipsilateral
shoulder and elbow markers. Movements made outside the horizontal plane were
not considered. For each joint angle, time series plots were aligned on movement
onset defined as the time at which the endpoint tangential velocity surpassed and
remained above 10% of the maximal value for that trial. Since initial positions
could vary between sessions, the absolute value of the angle at the final reach
position for each target was determined and used for comparison between
sessions. Trials were discarded if the child failed to complete the reach or dropped
the block (~20% trials). Finally, 4) trunk displacement was measured in mm as
sagittal displacement of the sternal marker between movement onset and offset
(time of grasping). Since the targets were aligned to the child’s midline, trunk
rotation was not assessed.

Statistical Analysis

Changes in clinical and kinematic data were determined for each child. Changes in QUEST scores were considered to be statistically significant according to the standard deviation (SD) band method (Trahan and Malouin 2002). This method consists of computing the mean of the baseline data (pre-tests 1 and 2). Then, a horizontal band representing ±2SD of the mean baseline and extending into the experimental phase (therapy + cast) and follow-up is traced. Data points measured in the post-test and follow-up falling outside the band were considered significantly different. The chance of a data point occurring outside the band without real change taking place is less than 5% (p<0.05). One-way within-subject repeated measures ANOVAs with factor time, were used to identify the effect of mCIT on each kinematic variable and post-hoc LSD (least significant differences) tests identified loci of significance at the p<0.05 level.
3.4. Results

**QUEST**

On initial testing, Jason had a QUEST score of 69.4 for pre-test 1 and 78.0 for pre-test 2 (Fig. 2A). After mCIT the QUEST score increased by 19.5% to 82.9, but this was not significant. Linda’s initial QUEST scores were 73.0 and 79 (Fig. 2B). After OT with left arm constraint (mCIT), the QUEST score increased by 23.7% to 90.3 and remained elevated at follow-up. Beth’s initial QUEST scores were 76.5 and 85.3 (Fig. 2C). The score increased significantly only at follow-up with an improvement of 26%

**Endpoint trajectory**

Jason’s trajectory smoothness did not change after OT alone or OT combined with mCIT for reaches within arm’s length (T1: $F_{3,8}=0.330$, $p>0.05$; T2: $F_{3,7}=0.122$, $p>0.05$; Fig. 3A). However, compared to pre-test 1, for reaches beyond arm’s length (T3), his trajectory become more segmented after OT alone as shown by the increase in the number of peaks at pre-test 2 ($F_{3,7}= 8.67$, $p<0.05$, Fig. 3A). For Linda and Beth, intensive OT alone and mCIT had no effect on hand trajectory smoothness for any target (Fig. 3C and 3E).

Jason and Linda did not change trajectory straightness after mCIT, although Jason tended to make straighter movements to T1 (Fig. 3B). Beth made more curved trajectories after OT alone but trajectory straightness returned to baseline values at the one month follow-up ($F_{3,28}=5.81$, $p<0.01$) for reaches to T1 (Fig. 3F). The opposite effect on trajectory straightness occurred for reaches to T3 which became straighter at post-test and follow-up ($F_{3,30}=4.78$, $p<0.05$, T3; Fig. 3F).

**Arm angles**

Jason and Beth increased elbow extension angles at post-test for reaches to T3 ($F_{3,4}= 7.41$, $p<0.05$, Fig. 4A; $F_{3,24}= 4.28$, $p<0.05$, Fig. 4E), and this increase was maintained by Jason at follow-up. The elbow extension angles at the final position for reaches to T1 indicate that both Linda and Beth decreased elbow extension at pre-test 2 ($F_{3,11}=8.58$, $p<0.01$, Fig. 4C; $F_{3,22}=5.39$, $p<0.05$, Fig. 4E)
but increased elbow extension after mCIT (p<0.01, p<0.01). Linda’s elbow angles returned to lower than baseline at follow-up (p<0.05). For reaches to T3, both Linda and Beth decreased their elbow extension angles at the one-month follow-up (F_{3,18}= 3.61, p<0.05, Fig. 4C; F_{3,24} = 4.28, p<0.05, Fig. 4E).

Shoulder horizontal adduction angles showed the most variability in each of the three children. For example, Jason showed unexpected results for reaches to T1 and T2. Shoulder horizontal adduction decreased at pre-test 2 and post-test and returned to pre-test 1 values at the one month follow-up for reaches to T1 (F_{3,7}=7.55, p<0.05) and to T2 (F_{3,6}=15.85, p<0.01, Fig. 4B). For reaches to T3, Jason’s shoulder movement was variable and did not significantly change. Linda decreased her shoulder horizontal adduction angles for reaches to T1 at pre-test 2, but this range increased and remained elevated at post-test and follow-up (F_{3,11}=7.35, p<0.05, Fig. 4D). For her reaches to T2, changes in horizontal adduction showed the same general pattern but these were non-significant. The greatest changes were observed for reaches to T3, for which shoulder horizontal adduction angles increased significantly at post-test and follow-up (F_{3,17}=12.84, p<0.001, Fig. 4D). Beth increased shoulder horizontal adduction angles at post-test for reaches to T1 after mCIT but not after OT alone (F_{3,19}= 6.85, p<0.01 , Fig. 4F). For reaches to T2, shoulder horizontal adduction angles decreased after OT alone at pre-test 2, but at follow-up this range increased to values higher than all previous assessments (F_{3,24}=7.95, p<0.005, Fig. 4F). Surprisingly for reaches to T3, shoulder horizontal adduction angles decreased at post-test and remained smaller at follow-up (F_{3,24}=28.01, p<0.001, Fig. 4F).

Trunk Displacement
All three children increased trunk displacement after mCIT. Jason’s initial values at pre-test 1 for reaches to T1 and T2 were within the range observed for typically developing (TD) children of his age (20 to 50 mm – T2; Schneiberg et al. 2004). For reaches to T2 and T3, after OT alone and mCIT, trunk displacement tripled (96 to 213 mm – T2 and 56 to 181 mm – T3) returning to lower values at the one month follow-up test. The increase was only significant for T3 (F_{3,5}=13.39, p<0.05, Fig. 5A). Linda’s trunk displacement increased after mCIT for reaches to
T1 (F3,14=5.01, p<0.05, Fig. 5B). For T2, Linda’s initial values ranged from 45 to 70 mm, which is greater than that expected for TD children. For reaches to T3, her trunk displacement decreased after OT alone, increasing again at post-test (F3,20=6.09, p<0.01, Fig. 5B). Beth had low initial trunk displacement values. After mCIT, her trunk displacement increased at post-test and returned to initial values at follow-up (F3,20=6.55, p<0.01, Fig. 5C). Beth’s trunk displacement for reaches to T2 decreased with OT alone, but increased after mCIT before returning to initial values at follow-up (F3,17=7.27, p<0.01, Fig. 5C). For T3, trunk displacement decreased after OT alone, but returned to initial values after mCIT and follow-up (F3,23=15.22, p<0.001, Fig. 5C).

3.5. Discussion

We aimed to determine whether improvements in clinical arm motor impairment following mCIT could be attributed to changes in arm movement quality as determined by detailed kinematic analysis. Three weeks of mCIT affected each child differently (Table 1). Jason only improved elbow extension for reaches to the far target without any positive change in the clinical test score or other kinematic measures. Linda improved QUEST scores along with the range of elbow extension to T1 and of shoulder horizontal adduction to two targets. Beth improved on QUEST and on several kinematic variables. Clinical and kinematic changes however were accompanied in all 3 children by increased compensatory use of the trunk after OT alone or after mCIT (see Table 1 and Fig. 5).

The change in QUEST scores was considered significant in two of the children based on the standard-deviation band method. These changes represented increases in QUEST scores between 23 and 26%, which is greater than the 3.2% considered to reflect clinically relevant improvement (Naylor and Bower 2005). These results add to growing evidence that mCIT has positive effects on clinical outcomes based on changes in clinical test scores. Our data suggest that while some clinical improvement may be due to improvements in reaching movement patterns, some improvement may also be related to an increase in motor compensations in the form of trunk displacement.
Jason did not improve in movement quality after mCIT aside from an increase in elbow extension for reaches to T3. While an increase in elbow range would be considered an indicator of improvement in movement quality, this was not accompanied by similar changes in trajectory parameters or shoulder range. In addition, these changes, combined with the increase in trunk displacement suggest that mCIT did not lead to an increase in arm movement quality.

For Linda, an increase in trunk displacement also occurred for reaches to T1 and T3 which was accompanied by increased shoulder horizontal adduction at post-test and follow-up and decreased elbow extension at follow-up for these two targets. The increase in trunk displacement may have been related to the decrease in elbow extension. In any case, the use of compensatory trunk movement did not result in any improvement in trajectory features.

Beth improved several kinematic characteristics in addition to the QUEST score. Except for shoulder horizontal adduction to T3, movement kinematics changed towards those observed in TD children of similar age (Schneiberg et al. 2002). However, once again, these changes were accompanied by increased trunk displacement. Beth initially used slightly more anterior trunk displacement for T2 than TD children (~50 mm). The amount of trunk displacement decreased after OT alone but increased significantly following the intervention (~75 mm). In this child, however, the effect was transitory with trunk displacement returning to more typical values at the one month follow-up. The improvement in trajectory straightness for reaches for T3 may be attributed to the increased trunk displacement and not to enhanced arm movement quality since there was no concomitant improvement in trajectory smoothness or shoulder movement. As has been previously shown in adults, it is likely that the additional trunk displacement was responsible, in part, for the straighter hand trajectory (Michaelsen et al. 2001).

The improvements in QUEST for Child 2 and 3 suggest that clinical improvements may be related to both improvements in some arm kinematic characteristics and to greater motor compensations. Trunk anterior displacement is a common compensatory movement used by adults and children with
hemiparesis. In adults, excessive trunk displacement may be used for arm transport during arm swinging, for reaching and for hand orientation during grasping.

Several explanations for the appearance of motor compensations after mCIT may be proposed. One possibility is that the child may not be able to make appropriate or coordinated joint rotations with the more affected arm to minimize trunk involvement due to the lack of maturation of cortical areas involved in sensorimotor integration (Paus et al. 1999). Several studies of motor learning in adult stroke survivors have shown that CNS injury impairs the ability to use sensorimotor feedback to monitor and improve performance in repeated movement trials (Platz et al. 1994). Another explanation may be the absence of feedforward control during reaching so that trunk displacement is not adequately prevented when the centre of mass is projected forward with the reaching arm (Schmitz et al. 2002). There may also be a delay in the development of perceptuo-motor function, particularly for visually-guided reaching in children with CP (Ferrel et al. 2001).

Reaching is a skill that improves with practice over the first decade of life (Schneiberg et al. 2002) involving a process by which the nervous system learns to select an appropriate motor strategy from a large repertoire of possible strategies (Hadders-Algra 2000). The reaching deficit in children with CP may reflect a more limited repertoire or choice in movement synergies (Hadders-Algra 2000), resulting on an incomplete separation of arm and trunk movement (Rossi et al. 2002). Indeed, animal studies have shown that the reaching movements of post-stroke rats, even after intensive rehabilitative therapy, were frequently repetitive and included combinations not observed prior to the stroke (Alaverdashvili et al. 2008). These studies suggest that such inappropriate gestures, termed “learned bad-use” substitute for and compete with successful movements. Thus, even when intensive movement therapy is provided during the period of limb constraint, the system will use the available degrees of freedom, which may not be the optimal ones, in order to accomplish hand and arm tasks.
Our data suggest that, without particular attention to limiting compensatory movements to extend the reach of the arm, constraint therapies may lead to the development of motor compensations, which may ultimately interfere with the learning of more optimal motor control patterns (Allred and Jones 2008; Alaverdashvili et al. 2008).

3.6. Limitations and Future Research

Few studies have demonstrated the effectiveness of interventions for motor improvement of the arm in children with CP. Previous studies of CIT in children for example, have demonstrated improvements in arm motor capacity or function, increases in range of joint motion and increased use of the arm (Eliasson et al. 2005, Charles et al. 2006) but none of these studies have used kinematic analysis to assess movement quality during performance of a functional task. Only detailed analysis of movement patterns can document the increased use of motor compensations that may accompany these changes. An obvious limitation of our study is that we only evaluated three children who had mild motor deficits due to CP so that the ability to generalize the findings to larger groups of patients is necessarily limited. Findings of decreased quality in movement patterns in all three children suggest that attention to motor compensations is warranted. Whether or not constraint therapies improve motor function via an increase in motor compensations should be addressed in future studies involving larger groups of children with CP.

3.7. Acknowledgements

We thank the children, their parents and occupational therapists from Marie Enfant Rehabilitation Centre who participated in this study. SS was supported by the Brazilian Government – Fundação de Coordenação de Aperfeiçoamento de Pessoal de Nível Superior (CAPES). MFL holds a Canada Research Chair in Motor Control and Rehabilitation.
3.8. References


Table 1: Summary of the effect of mCIT on clinical score and kinematic variables for each child.

Positive changes in movement quality (plus signs and hatched squares) are defined as higher QUEST score, lower number of peaks in endpoint trajectory (Trajectory smoothness), lower endpoint trajectory curvature (Trajectory straightness), increased range of joint motion (Elbow and Shoulder) and smaller trunk displacement. Minus signs and grey squares indicate negative changes.

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<th>Target/Child</th>
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<th>Trajectory smoothness</th>
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<th>Shoulder horizontal adduction range</th>
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Fig. 1: Study design, orthosis used to constrain the more-affected arm, schematic diagram of the reach-to-grasp task.

(A) Study design showing interventions and testing timeline; (B) Illustration of the removable bi-valve orthosis used to constrain the more-affected arm in children with CP; (C) schematic diagram of the reach-to-grasp task used for kinematic analysis. Objects were placed at three distances (T1–2/3 of arm’s length, T2–arm’s length, and T3-1 2/3 arm’s length) from the child’s body. T2 was placed at the limit of the child’s reach.
Fig. 2: QUEST scores.

(A) Child 1 – *Jason*, (B) Child 2 – *Linda* and (C) Child 3 – *Beth* at pre-test 1, pre-test 2, post-test and follow-up assessment sessions. Horizontal dashed lines indicate the mean ± 2 SDs of the two pre-test QUEST scores.
Fig. 3: Results for endpoint trajectory.

Histograms of mean (SD) trajectory smoothness (number of peaks - A,C,E), and straightness (Index of Curvature IC, B, D, F). Data are shown for reaches to 3 targets (T1, T2, T3) at each time period (pre-test1, pre-test2, post-test, follow-up). Significance indicated by asterisks *=p<0.05, **=p<0.01. Horizontal lines above bars in A and F identify the loci of the statistical differences.
Fig. 4: Results for arm angles.

Histograms of mean (SD) elbow extension (A,C,E) and shoulder horizontal adduction (B,D,F) for reaches to 3 targets (T1, T2, T3) at each time period (pre-test1, pre-test2, post-test, follow-up). Significance indicated by asterisks * = p < 0.05; ** = p < 0.01; *** = p < 0.001. Horizontal lines above bars identify the loci of the statistical differences.

Arm angles

Elbow extension

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Shoulder horizontal adduction

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Legend:
- Pre 1
- Pre 2
- Post
- Follow-up
Fig. 5: Results for trunk displacement.

Histograms of mean (SD) forward trunk displacement for reaches to 3 targets (T1, T2, T3) at each time period (pre-test1, pre-test2, post-test, follow-up). Significance indicated by asterisks *=p<0.05; **=p<0.01.; ***=p<0.001. Horizontal lines above bars identify the loci of the statistical differences.
CHAPTER 4

INTRODUCTION

From the results of the first study it was indicated that movement patterns in young children with CP detected by kinematic measures had high inter and intra variability. In addition, kinematic measurements were more sensitive to change after UL intervention than the QUEST clinical scale. For example, kinematic assessment revealed deterioration in movement coordination of two of the three children after mCIT (as manifested by a significant increase in trunk displacement) and the QUEST assessments rated these individuals as having improved their quality of movement.

In order to determine the most reliable kinematic variables for use in clinical trials, in this second manuscript we measured the test-retest reliability of kinematic measures of functional reaching movements in children with CP. The test-retest reliability was determined with intraclass coefficient model (ICC; 2, k). Thirteen children were recruited and assessed three times over an interval of five weeks length. The task measured kinematically was the same functional reaching task used in the previous study.
RELIABILITY OF KINEMATIC MEASURES OF FUNCTIONAL REACHING IN CHILDREN WITH CEREBRAL PALSY

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The study was conducted in Montreal, Quebec, Canada.

(Submitted to Dev Med Child Neurol)
4.1. Abstract
AIM. Dependable outcome measures that detect change need to be used to demonstrate upper limb (UL) rehabilitation effectiveness in children with cerebral palsy (CP). Thus, the reliability of kinematic measures of functional reaching movements in children with CP was evaluated.

METHODS. Thirteen children with spastic CP were evaluated three times over five weeks. Reach-to-grasp kinematics of a 2 cm³ wooden block placed at three distances from the body were analyzed from positional (x,y,z) data. Reliability of six kinematic variables was tested: straightness and smoothness of endpoint trajectories, trunk displacement, elbow extension, shoulder horizontal adduction and shoulder flexion. Test–retest reliability was determined with intraclass correlation coefficients (ICC, model 2,K) and 95% confidence intervals.

RESULTS. ICCs for all kinematic variables for all targets indicated moderate (ICC>0.50) to excellent reliability (ICC>0.90), except for shoulder horizontal adduction angles for reaches to close and middle targets and shoulder flexion for reaches to the middle target. Test-retest reliability was related to the workspace and task requirements.

INTERPRETATION. Kinematic variables can be used as outcome measures in clinical trials to test the effectiveness of UL interventions on movement quality. Kinematic variables are task specific and hence, reliability should be interpreted in the context of task requirements.

Key Words: test-retest reliability, upper limb, outcomes, rehabilitation, motor control
4.2. Introduction

Cerebral palsy (CP) encompasses a large number of non-progressive impairment syndromes arising from brain lesions or abnormalities in early development (fetal or infant). As the definition implies, children with CP have a variety of symptoms spanning sensorimotor, cognitive and social domains. In CP, brain injury occurring during periods of high neuronal plasticity and adaptability when projections from damaged CNS areas have not yet reached their final targets may interfere with essential processes of neural maturation. Therefore, while early neuronal organization facilitates motor skill acquisition (adaptive plasticity), this same capacity may lead to the development of atypical or alternative movement synergies interfering with typical development (maladaptive plasticity) as is often seen in children with CP. Furthermore, atypical movement strategies or motor compensations may be reinforced with practice and empower an individual because they result in successful task achievement yet interfere with the acquisition of more desirable movement patterns and mask real deficits. For example, emerging maladaptive movements may, over time, result in compromised posture, joint patency and range of motion (‘learned bad use’).

Judgment of therapeutic effectiveness in reducing CP symptomatology is directly linked to the outcome measures chosen to evaluate change. Thus, outcome measures should reflect the specific disabilities targeted by the intervention to provide unequivocal answers concerning treatment effectiveness. However, many motor outcomes used in clinical trials take the form of checklists measuring whether or not a task is accomplished but not how well the task is performed, i.e., movement quality. In order to fully account for adaptive movements, evaluation of treatment effectiveness should include both assessments of successful motor performance and movement quality. For the upper limb (UL), clinical scales such as the Quality of Upper Extremity Skills Test (QUEST) and the Melbourne Assessment of Unilateral Upper Limb Function permit subjective assessment of some elements of movement quality using continuous scales. Information about UL impairment gained from these
instruments may be more useful if complemented with more objective measures of movement quality such as kinematic analysis.

Kinematic analysis provides detailed quantitative data about movement patterns that can help an investigator relate changes in goal attainment to quantifiable changes in movement quality. For example, kinematic gait analysis is the gold standard for lower limb intervention effectiveness in children with CP \(^{14}\). However, kinematic analysis of UL movement in children with CP has received less attention \(^{15,16}\) possibly because of uncertainty of its reliability in this population. Indeed, a recent study examining hand movement between the knee, head and mouth in children with hemiplegia indicated low to moderate-to-high reliability of UL kinematic measures based on multiple correlation coefficients \(^{17}\). Thus, before considering assessments of UL interventions using kinematics, it is important to determine which measures are reliable kinematic indicators. Our goal was to identify kinematic variables that could be used as reliable outcome measures in future randomized clinical trials (RCTs) of UL reaching and manipulation interventions in children with CP.

### 4.3. Patients and Methods

Study participants were 13 children with spastic CP aged 6-11 years who had sensorimotor impairments in at least one arm, were able to sit unsupported, and had cognitive skills sufficient to understand instructions in English or French. Children with disorders such as ataxia, chorea, pain or orthopedic problems affecting the arm, neck or trunk including elbow or shoulder contractures greater than 10° were excluded. Children were recruited from five Quebec rehabilitation centers (MacKay Centre, Centre de réadaptation Marie-Enfant, Jewish Rehabilitation Hospital, Shriners Hospital, and Centre de réadaptation La ResSource). The study was approved by IRBs of McGill University, Ste Justine Hospital and the Centre de recherche interdisciplinaire en réadaptation (CRIR). The level of UL functional severity was classified with the Manual Ability
Classification System for Children with Cerebral Palsy – MACS (Table 2) - a 5 level ordinal scale classifying the ability to manipulate objects during daily activities of children aged 4-18 years. A MACS level 1 score indicates that the child handles objects easily and successfully and level 5 indicates that the child cannot handle objects at all and is severely limited in the performance of even simple actions.\(^{18}\)

### 4.4. Functional reaching task

To characterize reaching movement during a functional activity, a simulated feeding task was used. Children sat on a chair with their feet supported on the floor or a bench in front of a table adjusted to the height of the child’s elbow. In the initial position, before beginning each trial of the functional reaching task, efforts were made to maintain the child’s hand 5cm from the chest at sternal height, with the fingers straight, the index finger aligned with body midline, the thumb slightly abducted, the wrist and shoulder in neutral positions, the elbow flexed about 90°, and the forearm pronated. The same set-up was used for typically developing (TD) children in previous studies.\(^{25,26}\) Children were instructed to reach and grasp a 2cm\(^3\) wooden block at a verbal cue and to bring it toward the mouth as if eating a piece of food. Movements were self-paced and made with the more affected arm. The block was placed on target positions located on the table at three distances from the body midline (close target - T1: 2/3; middle target - T2: 1; far target - T3: 1 2/3 times arm’s length; Fig. 6). Target distances were based on the child’s arm length, defined as the distance from the medial border of the axilla to the distal wrist crease with the elbow extended.

Children were evaluated three times over 5 weeks (assessment 1 = 0wk, assessment 2 = 2.5wks; assessment 3 = 5wks) by the same evaluator. During this period, children did not receive any physical rehabilitation interventions. In each assessment, kinematic data were recorded with an Optotrak 3020 (Northern Digital, Waterloo) or a Vicon motion analysis system (Los Angeles, CA) at 100Hz. Data from 30 trials were collected in blocks of 10 trials per target, presented randomly. Trials were discarded if the child had trouble grasping or dropped the object, which occurred in <2% of trials. Infrared light emitting diodes
(IRED) or reflective markers were placed on anatomical landmarks and reference points of the arm and trunk: index finger middle phalanx (arm endpoint), thumb proximal phalanx, distal end of the 2nd metacarpal, radial styloid process, lateral epicondyle of elbow, ipsilateral acromion, sternal manubrium (trunk endpoint), contra-lateral acromion, and lateral to the ipsilateral iliac crest. Marker positions were standardized across assessments to improve reliability.

4.5. Data Analysis

Only the kinematics of the reach-to-grasp part of the task were analyzed. Positional (x,y,z) data were low-pass filtered (cutoff, 10Hz) and used to plot 3-D trajectories. Endpoint and trunk tangential velocities were computed from the magnitude of the velocity vector obtained by differentiation of positional data. Endpoint velocity traces were used to determine movement beginning and end. Movement beginning and end were defined as the times at which tangential velocities exceeded and remained above, or fell and remained below 5% of the maximal velocity. Movement end corresponded to the time of grasping the block.

Reliability of kinematic variables previously reported to characterize mature reaching patterns in TD children was tested\textsuperscript{19,20}: endpoint trajectory straightness and smoothness, trunk displacement, elbow absolute angle, shoulder horizontal (horizontal abduction/adduction) and sagittal plane movement (flexion/extension). Endpoint trajectory straightness was determined with the index of curvature (IC), defined as the ratio of the actual endpoint path length to that of a straight line joining initial and final positions, where a straight line and semi-circle have indices of 1 and 1.57 respectively. Endpoint trajectory smoothness was measured as the number of peaks (movement units) in the endpoint tangential velocity. A movement unit was defined as a local maximum velocity preceded and followed by increasing and decreasing values respectively for at least 20ms\textsuperscript{21} (see Fig.7). Trunk movement was computed as forward (sagittal) displacement in mm of the sternal marker (Fig.6). Final arm joint angles at the time of object grasping (movement end) were measured. Elbow and shoulder angles were computed by vectors joining the respective markers placed
on the wrist, elbow, ipsilateral shoulder and lateral iliac crest. The elbow angle was formed by the vectors between markers placed on the wrist, lateral epicondyle and ipsilateral acromion process where the fully extended arm was 180º. Shoulder horizontal abduction/adduction was measured as the horizontal projection of the angle between vectors formed by the ipsilateral and contralateral shoulder markers and the ipsilateral shoulder and elbow markers where 0º corresponded to the position in which the arm was outstretched directly in front of the shoulder in the frontal plane. Shoulder flexion was calculated with vectors formed by markers placed on the lateral epicondyle, ipsilateral acromion, and lateral iliac crest, where 0º was defined as the arm alongside the body.

4.6. Statistical analysis

The sample size of 9 subjects was estimated taking as the null hypothesis (ρ=0) the value of 0.70 representing moderate reliability as compared to the alternative or tested hypothesis (ρ=1) of 0.90 representing excellent reliability. Descriptive statistics (mean, SD) characterized kinematic variables for each child, assessment and target. To determine test–retest reliability, an intraclass correlation coefficient (ICC) model (2, K) and 95% confidence intervals (CI) were used. ICCs were based on two-way random effects ANOVA. This model was judged as most appropriate since it accounts for the random effects of subjects, average rating of the dependent variable at each time period, residual effects, as well as the number of observations.

4.7. Results

Effects of target distance on reaching performance

Typical endpoint trajectories in two children with different levels of severity in UL function (MACS scores 4 and 2) and their corresponding endpoint velocity and joint angle profiles are shown in Fig. 7. Targets required different amounts of arm and trunk displacement (Table 3). Trunk displacement and arm angles were smaller for T1 (close target) reaches than for the other targets. Endpoint trajectories were more curved to orient the hand in a frontal plane for
grasping the closer object. Reaches to T2 (middle target) did not require trunk displacement in TD children since the object was placed within arm’s length\(^\text{19}\). However, in the children with CP, the amount of trunk displacement and arm movement for T2 depended on the severity of the arm impairment of each child. Grasping T3 (far target) required trunk displacement and larger arm joint excursions. T3 endpoint trajectories were straighter than those for T1 and T2 since grasping this target required a more sagittal hand orientation. Shoulder horizontal abduction/adduction angles were small and relatively similar across targets because of the midline object placement (Table 3).

**Test-retest reliability in reach-to-grasp kinematics**

Reliability coefficients below 0.50, between 0.50 and 0.75, between 0.75 and 0.90, and above 0.90 represent poor, moderate, good and excellent reliability respectively (40). For our data, ICCs for all kinematic variables for all three targets had moderate (ICC > 0.50), to excellent reliability (ICC > 0.90), except for shoulder horizontal adduction for T1 and shoulder flexion for T2 (Table 3).

For reaches to T1, endpoint trajectory straightness (ICC = 0.59) and shoulder flexion (ICC = 0.60) had moderate reliability. Trajectory smoothness, and elbow angle had good reliability (ICC = 0.81, ICC = 0.86) respectively. Trunk displacement had excellent reliability (ICC = 0.92). Shoulder horizontal adduction had low reliability (ICC = 0.19).

For reaches to T2, trunk displacement (ICC = 0.95) and elbow angles (ICC = 0.94) had excellent reliability. Trajectory straightness (ICC = 0.76) and smoothness reliability (ICC = 0.88) were good. Shoulder horizontal adduction angle had moderate reliability (ICC = 0.50) and shoulder flexion angle had low reliability (ICC = 0.38).

For reaches to T3, trajectory smoothness, elbow angle, and shoulder flexion angle had excellent reliability (ICC = 0.91, ICC = 0.91, ICC = 0.93).
respectively. Trunk displacement had good reliability (ICC = 0.86) and shoulder horizontal adduction had moderate reliability (ICC = 0.51).

ICC Confidence Intervals (CI) were generally small for those variables with good to excellent reliability (Table 3, Fig. 8). Trunk displacement and elbow angles were the kinematic variables with the highest test-retest reliability coefficients and smallest CIs (95%) (Fig.8).

4.8. Discussion

Determination of UL rehabilitation effectiveness requires the precise measurement of change in movement patterns as well as their deviation from typical movement patterns. Therefore, it is essential to use measures that are able to detect the true variance inherent in the movement while still being reproducible. We investigated the reliability of kinematic variables describing reaching movement using ICCs and 95% CIs. The variables that rated consistently high across the three targets were trajectory smoothness, trunk displacement and elbow extension. Shoulder ab/adduction was not reliable at all, while trajectory straightness was reliable for reaches to both farther targets (T2, T3) and shoulder flexion was only reliable for reaches to the far target. As stated in the methodology, the advantage of using ICCs over correlation analysis (i.e. Pearson) is that ICCs account for the agreement between scores while correlation analysis is a bivariate measure of the relationship between two independent variables. The r coefficient in correlation analysis estimates how one variable will respond when a second variable changes and may be influenced by systematic changes in scores due to learning and practice effects. However, we used the ICC (2,K) model where "K" is the consistency definition and refers to the calculation of the ICC from an average of repeated trials. These ICCs are valid when a number of trials are used and an average taken.
Kinematic analysis: an unbiased measurement of functional reaching movement in children with CP

Most kinematic variables evaluated had good reliability with ICCs above 0.75. The acceptable level of reliability depends on the measurement goal. If the purpose of the measurement is to describe movement behavior, a lower reliability may be tolerated, especially if the source of the random variance is known. However, if the purpose is clinical decision-making or the demonstration of intervention effectiveness, reliability scores should be higher than 0.90\textsuperscript{24}. Our goal was to identify kinematic variables that could be used as reliable outcome measures in randomized clinical trials (RCTs) of UL interventions in children with CP. We took a less conservative approach and accepted reliability of .80 for three reasons. First, this study provides proof-of-principle of the feasibility of using kinematic variables for assessment and measurement. Second, movements in children with CP are highly variable and distinct from those of TD children. This suggests the need for an assessment approach that will appropriately accommodate alternate movement patterns. Third, a comprehensive analysis of the psychometric properties will require data from a substantially larger sample. The variables that rated consistently above an ICC of .80 across the three targets were trajectory smoothness, trunk displacement and elbow extension. Thus we are confident that these three parameters will be reliable indicators of change in movement quality for an UL reaching intervention.

Only one previous study assessed the test-retest reliability of UL kinematics in children with CP\textsuperscript{17}. They assessed the within-session (internal consistency) and the between-session reliability (test-retest – 1 week apart) of mean proximal and distal arm angle waveforms of two UL tasks performed by the more- and less-affected arms. Moderate \(r^2\) values were obtained for between-session reliability for measures of the affected arm, possibly due to differences in initial arm position. In our study, we had similar difficulties in controlling the initial position specifically of the shoulder which invalidated the analysis of change in range of joint angle data. Thus, we only reported final angular
positions. The differences in the task and type of statistical analysis in our and the previous study do not allow further comparisons.

**Test-retest reliability is specific to the workspace**

The reaching task was chosen to address UL movement deficits related to problems in self-feeding\(^{25,26}\). Three targets in the sagittal plane were used to investigate the effect of target distance on kinematic variables. Reliability coefficients varied according to the target distance. The lowest ICCs were computed for reaches to the nearest target, T1, and for shoulder horizontal abduction/adduction angles which had small values for all targets. The low ICCs may be explained by task-specific neural-biomechanical constraints\(^{27}\). For T1, reaching movements were shorter, visual gaze was different, and smaller trunk displacement and arm joint rotations were required than for the other target distances. These factors may result in small changes in environmental conditions and sensory information that may affect motor behavior in children with CP\(^{28}\). In addition, the relatively higher reliability for kinematics of reaching to T2 and T3 may have been related to a more limited number of arm and trunk configurations used for reaches to the farther targets. It is important to note that during reach-to-grasp movements from sitting, children with CP use more trunk displacement than their TD peers\(^{29}\). As a decrease of excessive trunk displacement is associated with better UL activity\(^{30}\), the high reliability of trunk displacement will permit analysis of this parameter in intervention studies.

Shoulder angle measures were not as reliable as those of the elbow as reflected in a higher between-measures variance (e.g., for T1: shoulder adduction=218, elbow=100). The ICC represents the ratio of the variance to the total variance (between subjects + between tests). If the variability among the repeated tests is smaller than the variability between subjects, the reliability will be good and the ICC will be high.

Finally, shoulder kinematic reliability may have been low because of the larger number of shoulder degrees of freedom leading to greater variability in
shoulder movement patterns used for reaching. There were three degrees of freedom at the shoulder (horizontal abduction/adduction, flexion/extension, rotation) compared to only one at the elbow (flexion/extension). Children had to reach and grasp the midline object. Since the hand initially was also in the body midline, the movement was done primarily in the sagittal plane, which required a combination of elbow extension and shoulder flexion. Shoulder movements in the horizontal plane (shoulder horizontal abduction/adduction) were small and contributed little to the task. Thus, the shoulder flexion measure was more reliable than that of shoulder abduction/adduction. Although the between-subjects variance of shoulder flexion and abduction/adduction measures was similar, the ICC for shoulder flexion was higher than that of shoulder abduction/adduction. The higher ICC was attributed to a lower between-measures mean square measurement error for shoulder flexion compared to that of shoulder adduction (e.g. root-mean square error for shoulder flexion to T1 was 16.94 compared to 218.26 for shoulder adduction). Thus, kinematic outcomes for the shoulder in the horizontal plane for UL interventions should be interpreted with caution. Further research is needed to address the test-retest reliability of shoulder horizontal angles for different UL tasks performed in different areas of the arm workspace.

4.9. Limitations

Kinematic variables are task specific so that reliability should be interpreted in the context of task requirements. Measurement reliability reported in this study can be generalized only for reaching and grasping objects placed in the body midline. In conclusion, kinematic analysis is a useful tool to investigate UL movement through objective description of movement quality for a specific task. Reliable kinematic variables can be used as main outcome measures in clinical trials aimed at evaluating the efficacy of UL interventions.
4.10. Acknowledgements

We thank the children and their families who participated in this study, Valeri Goussev for analytical programs, Gevorg Chilingaryan for statistical analysis advice, Jennifer Ranallo and Leah Feldman for data analysis. This study was supported by the Canadian Institutes of Health and Research (CIHR). SS supported by Fundação de Coordenação de Aperfeiçoamento de Pessoal de Nível Superior (CAPES). MFL holds a Canada Research Chair in Motor Control and Rehabilitation.
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Table 2: Demographic and clinical characteristics of the children in the reliability of kinematics study.

<table>
<thead>
<tr>
<th>Child</th>
<th>Sex</th>
<th>Age (years)</th>
<th>Impairment distribution</th>
<th>Most Affected UL</th>
<th>MACS*</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>M</td>
<td>11</td>
<td>Hemiplegia</td>
<td>Right</td>
<td>4</td>
</tr>
<tr>
<td>2</td>
<td>F</td>
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<td>Hemiplegia</td>
<td>Right</td>
<td>3</td>
</tr>
<tr>
<td>3</td>
<td>F</td>
<td>8</td>
<td>Quadriplegia</td>
<td>Left</td>
<td>4</td>
</tr>
<tr>
<td>4</td>
<td>M</td>
<td>10</td>
<td>Hemiplegia</td>
<td>Left</td>
<td>2</td>
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<tr>
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<td>F</td>
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<td>Right</td>
<td>3</td>
</tr>
<tr>
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<td>4</td>
</tr>
<tr>
<td>7</td>
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<td>Quadriplegia</td>
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<tr>
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<td>M</td>
<td>11</td>
<td>Hemiplegia</td>
<td>Left</td>
<td>2</td>
</tr>
<tr>
<td>Mean</td>
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<td>(9)</td>
<td></td>
<td></td>
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</tr>
<tr>
<td>(SD)</td>
<td></td>
<td>(1.6)</td>
<td></td>
<td></td>
<td>(0.9)</td>
</tr>
</tbody>
</table>

* Manual Ability Classification System for Children with Cerebral Palsy.
Table 3: Results for the test-retest reliability of kinematic data.

Test-retest reliability of kinematic data for reaches to targets placed at three different distances in three separate test periods for 13 children (Mean ± SD). ICC: Intraclass correlation coefficient and 95% confidence intervals (CI).

<table>
<thead>
<tr>
<th>Kinematic variables</th>
<th>Target 1 Mean (SD)</th>
<th>Target 2 Mean (SD)</th>
<th>Target 3 Mean (SD)</th>
<th>ICC (95% CI)</th>
</tr>
</thead>
<tbody>
<tr>
<td>N = 13 Children</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Trajectory Straightness (IC)</td>
<td>1.66 (0.44)</td>
<td>1.56 (0.19)</td>
<td>1.54 (0.23)</td>
<td>0.59 (-0.04, 0.86)</td>
</tr>
<tr>
<td>Trajectory Smoothness (# Peaks)</td>
<td>3.4 (1.3)</td>
<td>3.8 (1.0)</td>
<td>3.2 (0.9)</td>
<td>0.81 (0.51, 0.94)</td>
</tr>
<tr>
<td>Trunk displacement (mm)</td>
<td>26 (31)</td>
<td>35 (49)</td>
<td>39 (42)</td>
<td>0.92 (0.80, 0.97)</td>
</tr>
<tr>
<td>Elbow angle (degree)</td>
<td>72 (19)</td>
<td>77 (19)</td>
<td>76 (20)</td>
<td>0.86 (0.64, 0.95)</td>
</tr>
<tr>
<td>Shoulder Horizontal Adduction (degree)</td>
<td>45 (14)</td>
<td>47 (21)</td>
<td>39 (16)</td>
<td>0.19 (-1.1, 0.73)</td>
</tr>
<tr>
<td>Shoulder Flexion (degree)</td>
<td>44 (13)</td>
<td>42 (8)</td>
<td>44 (8)</td>
<td>0.60 (-0.06, 0.87)</td>
</tr>
</tbody>
</table>
Fig. 6: Experimental set-up of the functional reaching task used for kinematic analysis.

From a top-down (left) and sagittal (right) view with definition of parameters that were analysed. Objects were placed at three distances (T1–2/3 arm’s length, T2–arm’s length, and T3–1 2/3 arm’s length) from the child’s body. The task was to reach and grasp the object and bring it to the mouth region as in self-feeding. Only the reach-to-grasp component was analyzed.
Fig. 7: Kinematic variables.

(A) Mean endpoint trajectories of two children, with Manual Ability Classification System for Children with Cerebral Palsy (MACS) levels of 4 (left panels) and 2 (right panels), for reaches to close (T1 – thick solid lines), middle (T2 – thin solid lines) and far targets (T3 – dashed lines). (B) Mean endpoint tangential velocity profiles of the same two children for reaches to T2. A movement unit was defined as a local maximum velocity preceded and followed by increasing and decreasing values respectively for at least 20 ms (parallel lines). (C) Mean final angles of elbow extension and shoulder horizontal adduction for reaches to each target as defined in A.
Fig. 8: Intraclass correlation coefficients (ICC).

Intraclass correlation coefficients (ICC) for close (T1 - top), middle (T2-middle) and far targets (T3-bottom) for all kinematic variables: IC, index of curvature (endpoint trajectory straightness); # Peaks, number of peaks in the endpoint tangential velocity trace (endpoint trajectory smoothness); Trunk, trunk displacement; Elbow, elbow extension; ShHA, shoulder horizontal abduction/adduction; and ShF, shoulder flexion. Error bars are ICC ± 95% confidence intervals.
CHAPTER 5

INTRODUCTION

Findings from the first paper, where increased trunk displacement was an outcome for children with CP after receiving mCIT training, provide preliminary evidence that UL training practice can be accompanied by motor compensations. Moreover, from the review of the literature it was also revealed that to date no study investigated the effect of a task-oriented treatment approach on UL function in children with CP. Therefore, this third paper investigated the effect of a task oriented approach with trunk restraint (TR) strategy on UL movement quality in children with CP.

In order to use a measurement in clinical trials it is necessary that this measure is able to detect treatment effect changes. Therefore, kinematic variables used in this study are the ones determined in the second paper to have an ICC > 0.90 and include the following: hand trajectory smoothness, elbow, and trunk displacement. A clinical measurement that assesses motor compensations and has reported validity and reliability was selected, the Melbourne assessment.

In this study, the effect of task oriented training with TR was compared to the effect of task-oriented training without TR. The design of this study was a prospective single case research design (SSRD) with 12 children randomly allocated for each type of training. There were three baseline assessments in the, one assessment after the intervention, and a last assessment three months after the intervention.
THE EFFECT OF TASK-ORIENTED INTERVENTION AND TRUNK RESTRAINT ON UPPER LIMB MOVEMENT QUALITY IN CHILDREN WITH CEREBRAL PALSY – A RANDOMIZED N OF 1 TRIAL.

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\(^1\)Centre for Interdisciplinary Research in Rehabilitation (CRIR), \(^2\)School of Physical and Occupational Therapy, McGill University, \(^3\)University of Ottawa, \(^4\)Department of Epidemiology, Biostatistics and Occupational Health, McGill University.

The study was conducted in Montreal, Quebec, Canada.

(To be submitted to Pediatric Research)
5.1. Abstract

There is a lack of evidence about the efficacy of rehabilitation interventions to improve upper limb (UL) function in children with cerebral palsy (CP). The majority of the studies evaluating treatment interventions have not focused on the objective measurement of movement quality during task performance. Reaching, grasping and eating activities are impaired in children with CP. In addition, some studies have reported that reaching and grasping in adolescents with CP is followed by excessive trunk displacement, which can be considered as a motor compensation. The goal of this study was to investigate the effect of task-oriented training on UL function with and without a trunk restraint (TR) strategy on movement quality in children with CP.

The design of this study was a prospective single subject research design (SSRD). Twelve children were recruited, aged from 6 to 11 years. They were assessed three times before treatment, immediately after treatment, and three months after treatment completion. The main outcome measurements were kinematic variables during a functional reaching task and the Melbourne assessment. Children were randomly allocated to two types of treatment: task-oriented with TR strategy and task-oriented without TR strategy. The effect of both treatments was calculated with two typically approaches in SSRD: a regression line with all data observed at baseline phase, and standard mean difference.

The results suggested that task-oriented intervention with TR strategy promoted better improvements in UL movement quality in children with CP than task-oriented intervention without TR strategy. Although almost all children increased their elbow extension, only the children who received the task-oriented intervention with TR strategy were able to carry over their improvement three months after the intervention. Furthermore, some children who received the task-oriented intervention without TR doubled their pre-intervention trunk displacement levels.
5.2. Introduction

The term cerebral palsy (CP) embraces a variety of conditions that lead to impairments in sensorimotor functions (Baxter 2005). The updated definition states that the term CP describes a group of disorders affecting the development of movement and posture and causing activity limitation, which are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The new definition emphasizes that the motor disorders of CP are often accompanied by disturbances of sensation, cognition, communication, perception, and/or behaviour, and/or by seizure disorder (Bax et al. 2005). This updated definition provides a better description of the multidimensionality of the CP syndrome and the heterogeneity of its clinical features than previous ones (Mutch et al. 1992).

Motor impairments of the arm and hand are common in children with CP. The principal consequence of motor impairments in the upper limb is limitation in reaching, grasping and prehension activities (Henderson and Pehoski 2006), which are fundamental to self-care, education, and social activities. Children with CP can use alternative movement patterns during reaching and grasping compared to typically developing (TD) children (van der Heide et al. 2005; Hirschfeld 2007). The occurrence of lesions in an immature brain has severe consequences on motor development, especially when the brain injury occurs during the differentiation and connectivity (proliferation and pruning of synapses) phase of development (Johnston 2004). In addition, it has been demonstrated that during typical neurological development, afferent information and different experiences with upper limb movements may guide corticospinal connectivity with spinal cord circuitry (Eyre 2007). For example, in kittens, prevention of normal upper limb use during critical periods of corticospinal tract development led to prehension deficits (Martin et al. 2004). The typical development and maturation of spinal cord circuitry is essential for the transition from gross to more controlled, complex and fine movements. A lesion in an immature brain may result in aberrant ipsilateral corticospinal projections (Eyre et
which may lead to abnormal organization of spinal segmental mechanisms (Leonard et al. 1990; Jobin and Levin 2000). For example, in neonatal rats with unilateral brain lesions, the ipsilateral cortex may replace the function of the impaired corticospinal tract by formation of indirect pathways via the ipsilateral red nucleus (Z'Graggen et al. 2000). This post-lesional reorganization has been attributed to excessive or maladaptive plasticity (Johnston 2004). It has been suggested that the development of atypical or alternative movement synergies following early brain injury may be due to reorganization in the immature brain which may in turn interfere with typical development (Alaverdashvili et al. 2008a). In addition, motor compensations might be common in children with CP since the injured CNS is continuously adapting to find new movement solutions to a given motor problem (Thelen 1996). The appearance of motor compensations may be considered maladaptive because they may prevent children from experiencing normal movement patterns. Given the likelihood for children with CP to develop non-typical movement patterns during early development, therapies strive to improve movement quality to avoid the establishment of maladaptive behaviors i.e. ‘learned bad use’ (Carr and Shepherd 1996; Alaverdashvili et al. 2008a).

Studies of reaching, grasping and eating have reported that children and adolescents with CP use excessive trunk displacement for such activities in comparison to their non-disabled peers (van Roon et al. 2004; Kreulen et al. 2006; Coluccini et al. 2007; Kreulen et al. 2007). The excessive trunk movement observed in children with CP is considered to be abnormal because it can be twice the amount of trunk movement observed in TD children (van Roon et al. 2004; van Roon et al. 2005; Mackey et al. 2006). Positive results in reaching movement quality in children with CP as decreased in muscle co-contractions and increased in speed have been reported when the trunk movement was restrained by the therapist (Hirschfeld 2007) or by an external device (van der Heide et al. 2004; van Roon et al. 2005), or when the trunk was supported in good vertical alignment (Butler 1998; Stavness 2006). In one study in adult stroke survivors in which excessive trunk movement was restrained by straps during a five-week
upper limb treatment intervention, participants increased active shoulder and elbow range of motion during reach-to-grasp task and improved functional ability in the arm and hand (Michaelsen et al. 2006). The results of these intervention studies support the idea that excessive trunk displacement may be undesirable during the performance of some upper limb tasks.

There is a lack of evidence about the efficacy of rehabilitation interventions to improve UL function in children with CP (Boyd et al. 2001). This may be partly attributed to the small number of randomized clinical trials (RCTs) of good methodological quality (Anttila et al. 2008), and to the use of inadequate outcome measurements (Wasiak et al. 2004; Hoare et al. 2007). Studies evaluating treatment interventions have not focused on the objective measurement of movement quality during task performance so are unable to determine if post-intervention improvements were due to an increase in the use of motor compensations or to improvements in movement quality (Cirstea and Levin 2000; Levin et al. 2002; Alaverdashvili et al. 2008b).

Most of the outcome measures used in clinical trials to improve UL function in children with CP do not provide sufficient detail of movement quality (Charles et al. 2006; Deluca et al. 2006; Satila et al. 2006; Wallen et al. 2007). A more quantitative and objective way of investigating improvement in movement quality is with kinematic analysis. The advantage of kinematic analysis over subjective-observational outcome measurements is that it can provide detailed data about movement performance, which allows researchers to relate changes in goal attainment to improvements in movement quality.

A third explanation for the lack of high level evidence of UL therapeutic effectiveness in children with CP is that previous studies addressed impairments instead of function and activity (Damiano 2008). The concepts established by the International Classification of Functioning, Disability and Health (ICF) state that body functions, activity, environment and personal factors are inter-related (Rosenbaum and Stewart 2004). The task-oriented treatment approach in rehabilitation is based on the ICF model (Shumway-Cook and Woollacott 2007).
By developing efficient task-specific strategies and adapting goal-oriented movement to different tasks and environments, the therapist can provide a variety of conditions to practice UL function that will be activity (task) dependent and related to the main underlying impairments in tasks specifically designed for the individual (Sackett 2000; Shumway-Cook and Woollacott 2007). The aim of this study was to investigate the effect of task oriented training on UL movement quality in children with CP. Specifically, we investigated whether improvement in UL movement quality may be better when task oriented training was combined with a trunk restraint strategy. We hypothesized that task oriented training would improve UL movement quality in children with CP, and that improvements would be maintained at three months following the end of the intervention. We also hypothesized that a better effect on movement quality with a decrease in movement compensations would be detected in children who received task-oriented training with trunk restraint strategy compared to those who received task-oriented training alone.

5.3. Methods

5.4. Participants

Twelve children with CP were recruited between October 2005 and November 2007 from four pediatric centers in Montreal (MacKay Centre, Centre de réadaptation Marie-Enfant, Jewish Rehabilitation Hospital, and Shriners Hospital) and one in Gatineau (Centre réadaptation de La ResSource). The inclusion criteria to participate in the study were: having a diagnosis of spastic CP, being between 5 to 12 years old, having motor function deficits in at least one of the upper limbs, being able to sit without trunk support, and having a good understanding of either English or French. Reasons for exclusion were the presence of cognitive deficits, athetoid movements, ataxia or choreoathetosis, chronic pain or orthopedic problems affecting the arm, neck or trunk, contractures of more than 10 degrees in the elbow or shoulder joints, and if the CP had a
trauma etiology. All parents signed the consent form approved by the ethics committee of the Centre de recherche interdisciplinaire en réadaptation (CRIR), and the Institutional Review Boards of Ste Justine Hospital and McGill University. If children were eleven years old or more they also had to sign a child assent form.

5.5. Study design

This study is a single subject research design (SSRD) with a baseline, post-intervention and follow-up assessments (A-B-A). Because of the heterogeneity of children with CP, the use of a single subject research design (SSRD) combined with outcome measures sensitive to changes in movement patterns is recommended over group comparisons to investigate treatment effects (Siebes et al. 2002). Furthermore, SSRD when carefully conducted by respecting internal validity issues, can be considered an optimal design to implement scientific evidence to practice (Patrick et al. 2000). To date, only a few studies have used this methodology to evaluate treatment outcome in children with CP (Thorpe and Valvano 2002; Trahan and Malouin 2002; Shumway-Cook et al. 2003), while others confound SSRD design with case reports or use methods more appropriate for the group analysis. In our study, the baseline assessment (A1) consisted of three assessments performed during a 5-week period before the intervention. The post intervention assessment was performed just after or up to a maximum of one week after the intervention was completed (B). For the follow-up assessment (A2), the evaluation was scheduled 3 months after the post-intervention assessment.

5.6. Measurements

Screening and clinical characteristics

UL passive range of motion (PROM) and pain were measured with the Fugl-Meyer Scale, where a maximal score of 24 represents full range and no pain.
The same scale was used to evaluate light touch and proprioception, where a maximal score of 20 represents no impairment light touch perception and a maximal score of 8 represents no impairment in position sense. Perception of deep touch was assessed with the 2-point discrimination and Semmes-Weinstein tests (Lafayette Instrument) TD children aged from 5 to 9 years score between 2 mm and 3 mm for the 2-point discrimination test (Cooper et al. 1995), and for the Semmes-Weinstein test, values around 2.83 and 3.61 thresholds are considered normal (Bell-Krotoski et al. 1995). Spasticity was measured using the Composite Spasticity Index (Levin and Hui-Chan 1992), where a maximal score of 16 represents severe spasticity. The level of severity of UL function in each child was classified with the Manual Ability Classification System for Children with Cerebral Palsy (MACS). MACS is a 5 level ordinal scale used to classify the ability to manipulate objects in daily activities in children between the ages of 4 and 18 years (Eliasson et al. 2006). A MACS level 1 score means that the child handles objects easily and successfully and a score of level 5 indicates that the child cannot handle objects at all and is severely limited in the performance of even simple actions.

Main outcome measurements

A functional reach test was used to assess the ability of the child to reach and grasp a 2 cm3 block (target) in a simulated self-feeding task. Children were seated on a chair and their feet were supported on the floor or on a bench. The table height was adjusted to the height of the child’s elbow while he/she was sitting in the chair. For standardization, the object was placed in line with the child’s body midline at two target distances proportional to the child’s arm length (T1 - 2/3 of arm’s length, T2 – arm’s length). The arm’s length was defined as the distance from the medial border of the axilla to the distal wrist crease with the elbow maximally extended. The initial position of the tested hand was 5 cm from the chest at sternal height, fingers straight, index finger aligned with the body midline, thumb slight abducted, wrist and shoulder in neutral positions, elbow
flexed about 90°, and forearm pronated. The same experimental set up was used for TD children in a previous study (Schneiberg et al. 2002; Sveistrup et al. 2008). A block of 10 to 15 trials was recorded for each target and blocks were performed in a random order. Movements were made with the most affected arm and children were instructed to move at a self-paced speed. The functional reach test was performed without trunk restraint. The kinematic variables analyzed were smoothness of hand trajectory, elbow angle at the final position, and trunk displacement. The test-retest reliability of these variables were investigated in previous studies and their intraclass coefficients (ICCs) were greater than 0.90 (Schneiberg et al. (In press)).

The main clinical outcome used was the Melbourne Assessment of Unilateral Upper Limb Function (Johnson et al. 1994; Randall et al. 2001). The Melbourne is a valid and reliable evaluative tool that measures unilateral UL quality of movement in children from 5 to 15 years of age applicable to children challenged by any neurological dysfunction. UL impairment and movement compensations are rated during 16 specific UL activities. Intra-inter rater reliability of the scale is good (Kappa coefficient values greater than 0.87) (Randall et al. 2001). However, to date no information about its sensitivity and responsiveness to meaningful clinical change has been reported.

5.7. Randomization

After the screening assessments, each child was randomly allocated to receive the task-oriented intervention either with or without trunk restraint. The person responsible for treatment allocation was not involved in the recruitment, evaluation or treatment of the children, did not have any contact with the child or their parents, and did not have any information about the outcome of the clinical assessments. The only information provided to this individual was the child’s age and their MACS level. Allocation of treatment was concealed by private and confidential emails to therapists (Occupational and Physical Therapists – OT and PT). The therapists’ involvement in the project was exclusively to deliver the treatment.
5.8. Intervention

All children participated in a training intervention consisting of a 1 hour treatment session, 3 times per week for 5 weeks. The completed intervention therefore included 15 sessions with a total of 15 hours of treatment. The intervention was delivered by PTs or OTs in each pediatric center. The 1 hour intervention session was divided in 5 blocks. The first block included a warming up or preparatory activity lasting 3 minutes. In this block the therapist could stretch, and/or mobilize the child’s UL before starting the next activity. Each child sat in a height-appropriate chair during therapy. The chair height was chosen so that, while wearing shoes, the child’s feet could be firmly supported on the foot rest. The table height was adjusted to the child’s elbow height. The second block was the task-oriented practice lasting 20 minutes. The task-oriented block was practiced in a standardized environment within the confines of a custom-built wooden box in which different workspace zones were defined (Fig. 9). The box was built so that upper limb movements could be performed in a horizontal plane (e.g. ipsilateral, contralateral, close and far zones of reaching and grasping) and a vertical plane (e.g. high and low zones of reaching and grasping, reference point between high and low reaches was 90 degrees of shoulder flexion, horizontal adduction and abduction). The vertical wall of the box was painted with magnetic paint on which the child could place magnetic stickers. Several toys of different shapes, sizes and weights were used by therapists during the training. Toys were chosen based on clinical judgement of each child’s preferences and needs. Toys that were easy to manage were appropriate for children up to 3 years of age, while toys that were more difficult to manipulate were appropriate for older children (up to 15 years in age; e.g. Mr. Potato Head™-Playskool and Connect Four™). The task-oriented block included uni- and bimanual activities requiring arm movements in different parts of the workspace. The workspace, type of grasping, and size, weight and shape of the toys were chosen according to the individual child’s needs. After the task-oriented block, the child had a 7 min break, following which, the third block was done. This block consisted of task specific
activities done within a virtual reality (VR) game environment. The VR environment was a commercially available video-capture game platform (IREX, GestureTek, Toronto, ON). The IREX system was adapted to project the hand and forearm onto the screen instead of the whole body so that the UL of the child could interact with virtual objects in different games. In order to play the game, the child had to move their UL in different directions (Fig. 10). Therapists were encouraged to use clinical judgment while positioning and explaining the UL movements to the child. They emphasized movement in ranges that would challenge the child. All five pediatric centres were equipped with the wooden box, two baskets of toys, and one computer with a flat screen and webcam for the VR and the IREX program. The last block of the intervention was the functional training, lasting 10 minutes. In the functional training the child had to choose one UL activity that they wanted to perform better or that they could not perform. If the child was unable to choose an activity, the therapists asked the family to suggest one.

Trunk restraint

Children were randomly allocated to receive either task-oriented practice with (TR) or without trunk restraint (NTR). Children allocated to TR practice had their forward trunk displacement and trunk rotation movements limited by two Velcro straps that criss-crossed their chest and were attached to the chair (Fig. 11). The straps allowed an age-appropriate amount of trunk movement during the upper limb tasks. The amount of trunk movement permitted by the straps was based on previous data from TD children (5 yrs = 5 cm, 6-7 yrs = 4 cm, 8-9 yrs = 3 cm, 10-12 yrs = 2 cm; Fig. 11) (Schneiber et al. 2002). The trunk restraint did not limit movements of the shoulder girdle so that shoulder flexion, horizontal adduction and abduction, and shoulder rotation was not constrained. It should be noted that during the functional training block (10 min), none of the children had their trunk restraint in place. During this training block, the child and his/her family chose the activity to be practice (child-centred approach) (Law et al.
Since the trunk restraint straps may have limited the type of activities that could have been chosen by the child, no trunk restraint was used. In the NTR group, no verbal instruction regarding trunk use during upper limb activities was provided by the therapist, but therapists could guide arm movement or motivate the child as is usually done in routine clinical practice.

5.9. Blinding
Both the evaluators and children were blinded to treatment allocation. The evaluator responsible for collecting and analyzing the kinematic and clinical data was unaware of treatment allocation and the order of assessments when analyzing kinematic data or videotaped clinical evaluations. PTs evaluators responsible for scoring the Melbourne test received coded and randomized videotapes, each holding evaluations from four to five different children. The evaluators also did not have any contact with the child or the therapists involved in the intervention. Children were blind to treatment allocation because in both interventions, the trunk restraint straps were placed over the child’s body. For the TR training, the straps were attached to the chair limiting trunk displacement while for the NTR training, the straps were loose and did restrict trunk movement.

5.10. Data analysis
Kinematic data

Ten (10) markers (infrared emitting diodes = IREDs) were placed on strategic anatomical locations on the child’s body: tip of the index finger, second phalanx of the thumb, head of metacarpus of index finger, radius styloid process, middle of forearm, lateral epicondyle, ipsilateral acromion, manubrium sternum, contralateral acromion, and lateral iliac spine. Positional data (x, y, z) were low-pass filtered (cutoff, 10 Hz) and used to plot 3D trajectories. In each assessment, data were collected with Optotrak 3020 (Northern Digital, Waterloo) at a sampling rate of 100 Hz for 5 to 8 sec depending on the child. For analysis of trajectory smoothness, the number of movement units was determined as the number of peaks in the tangential velocity profile. A movement unit (MU) was
defined as a local maximum velocity preceded and followed by increasing or
decreasing values respectively for at least 20 ms (von Hofsten and Lindhagen
1979). The amount of elbow extension at the time of grasping (elbow angle) was
calculated by analysis of vectors formed between markers placed on the wrist and
lateral epicondyle and the lateral epicondyle and the ipsilateral acromion process.
The fully extended position of the arm was defined as 180° elbow extension.
Trunk displacement was computed as the distance moved by the marker on the
sternal manubrium in the forward direction. Seven to 13 trials per kinematic
variable were used in the analysis. However, a trial could be discarded if the child
had trouble grasping or dropped the object, or if missing marker data could not be
replaced. However, for some kinematic variables, up to 50% of the trials had to be
discarded, because of missing markers.

5.11. Statistical Analysis

To estimate the effect of the treatment intervention on the kinematic
outcomes in each child, two statistical approaches commonly used in SSRD were
used (Kazdin 1982; Barlow et al. 2009): a regression approach with a visual trend
analysis and a non-regression approach with the effect size obtained through the
standard mean difference. For the visual trend analysis, the first step was to
investigate autocorrelation using the Bartlett test (Payton 1994). Since no
dependency in baseline observations was found, all observation points made
during the three baseline assessments were combined. A linear regression line
was fitted through the baseline data points. A second straight line was fitted at the
end of the baseline regression line that went through post-intervention (B) and
follow-up assessment (A2). The observations above or below the second line
were counted for each assessment. To determine the effect size of the intervention
for each child, the difference was calculated between the mean of the post
intervention assessment (x^_B) and the mean of the baseline (x^_A1) over the
standard deviation (SD) of the baseline (Effect Size, E.S. = (x^_B - x^_A1) /
SDB). To determine the effect size of the follow-up assessment for each child, the
difference was calculated between the means of the follow-up ($\bar{x}_2$) and the baseline assessments ($\bar{x}_1$) divided by the standard deviation (SD) of the baseline, (Olive and Smith 2005). Effect sizes of 0.20, 0.50 and 0.80 were considered to be small, moderate and large according to Cohen (Portney and Watkins 2000).

To estimate the effect of the intervention on the Melbourne assessment, a 2SD band method was used (Trahan and Malouin 2002). This method consisted of computing the mean and SD of the baseline data. Then, a horizontal band representing $\pm 2SD$ of the mean baseline and extending into the intervention assessment (B) and follow-up (A2) was traced. Data points measured in the post-test and follow-up falling outside the band were considered significantly different from baseline assessments. The chance of a data point occurring outside the band without real change taking place was less than 5% ($p<0.05$).

5.12. Results

Clinical and demographic characteristics at baseline for each child are presented in the Table 4. All children completed all the study assessments and fully complied with the intervention sessions. One child missed one session because of illness. A replacement session was scheduled at the child’s convenience.

Overall the following observations were noted: a) changes in trajectory smoothness for reaches to both targets were similar among children involved in both types of practice; b) elbow extension improved following the intervention assessment for both distances, with deterioration at follow-up for reaches to T2 for those in the NTR practice; c) trunk displacement was improved at intervention and follow-up for reaches to T1 and at follow-up for reaches to T2 for children in who had the TR training, while this aspect deteriorated for children who had the NTR training at both intervention and at follow-up to reaches at T2. These are shown in the trend line graphs for all subjects (Figs. 12 and 13) in the summary of effect sizes for assessment B (Fig. 14) and in the tables of mean
differences and effect sizes for the hand trajectories (Table 5), elbow extension (Table 6) and trunk displacement (Table 7) reaching to the two targets.

Hand trajectories

Improvement in smoothness of hand trajectories was characterized by a decrease in the number MUs (data points below the baseline trend) and a deterioration was characterized by an increased number of MUs – data points above the baseline trend. Data points and trend lines in arm trajectory smoothness for all 12 children are shown in Figs. 12 and 13 (left columns) for reaches to T1 and T2, respectively. The numbers of points below the trend line for each child, target and time period are shown in Table 5. Improvement and deterioration in arm trajectory smoothness are also indicated in Table 5 as negative or positive effect sizes respectively.

In the TR group, overall for reaches to the close target (T1), there was a mixed response, with trajectories becoming smoother in four children after the intervention (Ch 3, Ch 5) or at follow-up (Ch1, Ch3, Ch6). The same four children (Ch1, Ch3, Ch5, Ch6) improved arm trajectory smoothness during reaches to the far target.

A similar effect was observed in children in the NTR group. Three children (Ch7, Ch8, Ch11) had smoother trajectories after the intervention or at follow-up for T1 reaches and four children (Ch8, Ch9, Ch11, Ch12) made smoother movements during reaches to T2.

Elbow angle

Improvement (increase) in active elbow extension was identified in children receiving both type of practice but this was more marked in those who had the TR intervention. As shown in Figs. 12A and 13A, second column, improvements in elbow extension were characterized by points above the baseline trend line. For reaches to T1, all children (Ch 1- 6) who received the TR intervention increased elbow extension angles and effect sizes were substantial.
for all but one child (range from 0.60-3.34) after the intervention (Table 6). Four of the six children maintained the improvement at follow-up (Ch 1, 3, 4, 6) while values returned to baseline in one child (Ch 2) and deteriorated in one child (Ch 5).

For the children who received the NTR intervention, four (Ch 7 -10) improved elbow extension after the intervention with large effect sizes (range 1.06 - 3.54) for reaches to both targets. Only one child (Ch 9) maintained the improvement in T1 reaches at follow-up and another child maintained the improvement at follow-up for T2 reaches (Ch 7). One other child (Ch 11) improved elbow extension to T1 and T2 only at follow-up. In contrast to the TR group, elbow extension ranges became worse in half the group (Ch 8, 10, 12) at follow up for reaches to T2. As in the TR group, elbow extension range decreased in one child (Ch12) for both targets and at both time periods (Table 3). Thus for the TR group, improvements that were observed in the children post-intervention were maintained at the follow-up period, while this was generally not the case for the NTR group.

Trunk displacement

Improvement in trunk displacement was defined as a decrease in trunk flexion during reaches to each target, identified by data points falling below the baseline trend (Figs. 12 & 13, 3rd column). For those who received the TR intervention, for reaches to T1, five of the six children improved trunk displacement with high effect sizes and all but one child (Ch3) used less trunk displacement at follow-up (Table 7). For reaches to T2, improvement was observed in only one child (Ch1) following the intervention and in this child and three others (Ch1, 2, 4, 5) at follow-up.

In contrast, for reaches to T1, only one child (Ch 11) in the NTR group improved (decreased) trunk displacement, with a large effect size after the intervention that continued to improve at follow-up (Table 7). In one child (Ch 9) trunk displacement increased after the intervention and at follow-up (effect size 1.26) while another child (Ch7) showed a deterioration with a moderate effect size at
follow-up. For reaches to T2, only one child (Ch11) decreased trunk displacement after the intervention but this was not maintained at follow-up. In contrast, in three children (Ch 7, 9, 10) trunk displacement increase after the intervention and remained high at follow-up (Table 7, Fig. 13B).

Thus overall, improvement in trunk displacement was better for the TR than the NTR at T1. For reaches to the farther target T2, the TR group showed delayed improvement at follow-up while the NTR group showed deterioration after the intervention that was maintained at follow-up.

The results of the Melbourne Assessment (Fig.15) showed that in the TR group, only Child 4 improved in both post-intervention and follow-up assessments and Child 7 improved only in the follow-up assessment. None of the children in the TR group lost movement quality as a result of the intervention. In the NTR group, one child (Ch 7) improved slightly at the follow-up evaluation. However, four children showed mild deterioration in the Melbourne Scale score, three of them following the intervention (Ch 7, 8, 10) and one at follow-up.

5.13. Discussion

The purpose of this study was to investigate the preliminary effect of task oriented intervention on UL movement quality measured by kinematic analysis and Melbourne clinical assessment in children with CP. Our specific objective was to examine if task oriented training combined with a trunk restraint strategy (TR) had a better effect than task oriented training alone (NTR). Our first hypothesis was that a task oriented intervention would improve aspects of UL movement quality and that improvement would remain during the three months following the end of the intervention. Our second hypothesis was that children who received task oriented training with TR would make greater improvements on UL movement quality due to a decrease in movement compensations than children who received task oriented alone (NTR).

The results of this study tend to support our hypotheses (Table 8). Smoothness of hand trajectory (# peaks) and elbow extension angles improved in
more than 50% of the children in both interventions. However, there were differences in the kinematic outcomes between the two groups that suggest that the task-oriented intervention with TR may be more effective. Improvements in elbow extension continued into the follow-up period for the TR but not the NTR subjects. More importantly, trunk displacement was only effectively reduced in children who received TR. Furthermore, three children (Ch 7, 9, 10) who received the NTR had a noticeable increase in trunk displacement after the intervention and at the three month follow-up assessment.

The Melbourne outcome measure did not yield meaningful results however, as only one child improved in this assessment (Ch 4) after the intervention. We chose the Melbourne because it is a reliable and valid tool that measures 16 UL activities. In addition, the Melbourne assessment accounts for movement compensation, range of motion and fluency of the movement, which are variables that characterize movement quality. The Melbourne is scored by videotape and is influenced by subjective judgments. However, while it has been reported that the level of the training can influence the inter-rater reliability of Melbourne (Cusick et al. 2005), we were careful to select experienced therapists who were skilled at using the tool. As there have not yet been any studies that have reported the sensitivity or responsiveness to change of the Melbourne assessment, it is possible that this instrument was not as sensitive as the kinematic analysis in capturing changes in movement quality. This indeed may be the case, as alluded to in other studies in children with CP assessing the effects of botulinum toxin A (Speth et al. 2005; Wallen et al. 2007).

The large effect size in kinematic parameters were due to small baseline variability combined with large post-intervention changes. A critical element in interpreting the large effect sizes is the reliability of the kinematic measures (Baugh 2002). Thus we used only those kinematic variables that were shown in a previous study with the same cohort to be reliable with an ICC of 0.9 or above (Portney and Watkins 2000). Therefore, we feel confident that the effect sizes are a dependable indication of the changes in movement performance.
Task-oriented approach to improve movement quality.

Characterization of functional reaching in TD children is related to the achievement of an adult kinematic reach and grasp profile (58). This includes the presence of a bell-shaped velocity profile representing a single movement unit by 6 - 7 years of age, with the degree of elbow extension and trunk displacement scaled to the reaching distance (Schneiberg et al. 2002). However, children with CP have problems with reaching and grasping which is often characterized by multiple peaks in the velocity profile of the hand movement as well as excessive trunk flexion and decreased elbow extension for a given reaching distance (Van Thiel and Steenbergen 2001; van der Heide et al. 2005). Our study has indicated that the quality of movement in these three parameters depended on the distance of the reaching target, the parameter measured and the task-oriented intervention (TR or NTR).

Hand Trajectories

The effect of the intervention on hand trajectory smoothness was not clear-cut, although for some children, there was a substantial improvement following the intervention, as manifested by a clear decrease in the number of MUs during the reach. In particular about 50% of the children in each group improved substantially for reaches to either one or both targets, following the intervention or at follow-up. As these were not always the same children, this skill may be one that is not easily learned and may require considerable practice or time to emerge. Indeed, trajectory smoothness in three children (Ch 2, 6, 12) deteriorated at the end of the intervention for reaches to T1. These children all used their more-affected left arm for reaching, they had low scores on position sense testing, and two of them had low stereognosis and light touch sensory scores (Table 4). In our previous studies on the development of coordination of reaching movements (Schneiberg et al. 2002) and development of head and trunk coordination in TD children (Sveistrup et al. 2008), a common finding was that development into mature patterns was acquired later for the reaches to the closest
target (T1). Those findings might suggest that reaches to objects placed close to
the body required a more complex sensory integration and trajectory formation.

Transcranial magnetic stimulation (TMS) and functional magnetic
resonance imaging (fMRI) studies in children with hemiplegic CP (Thickbroom et
al. 2001) have demonstrated abnormal ipsilateral hemispheric projections with
shorter latencies than contralateral projections. Furthermore, passive and active
movements activate the contralateral hemisphere, suggesting an inter-hemispheric
dissociation between afferent kinesthetic inputs and efferent corticomotor output.
Possible relationships between impairments, site, size of the lesion, and time of
injury have been reported (Thickbroom et al. 2001). In our study we did not have
information about the site of lesion in the children which limits us from
suggesting a relationship between the site of lesion and the findings. In addition,
another SSRD study that investigated the effect of balance training in children
with CP (Shumway-Cook et al. 2003) also found that responses to the training
varied according to the type of impairment of the child. It could be that children
with sensory deficits, especially position sense, need a different type of training
for reaching to objects close to the body.

Indeed, the meaning of the number of movement units (MUs) or peaks
in the velocity profile of reaching movements in hand trajectory formation has
been long been debated (von Hofsten and Lindhagen 1979; Fetters and Todd
1987; Jeannerod 1988). Some authors have suggested that the decrease of MUs
during development represents different solutions for dynamical and
biomechanical problems for each infant (Thelen et al. 1993; Thelen et al. 1996).
Others have suggested that movement units could be deliberate corrections to the
trajectory at a higher level of motor control. Thus, the segmented trajectory could
result from the infant’s inability to generate a virtual trajectory (Feldman and
Latash 1982; Feldman 2008). Thus, the type of impairment and previous
experience could be important factors in determining outcome in this parameter.

Elbow angle
Our study showed that elbow extension consistently improved across both target distances regardless of treatment intervention. The effect size for elbow extension after the intervention for both groups was large, ranging from 0.60 to 3.75, which reflects differences in elbow extension ranging from 10 to 47 degrees. This range of improvement in elbow movement may be clinically meaningful when considering the presence of flexion contractures in children’s elbow (Jandric 2007; Nobuta et al. 2008). Only one child in the NTR training decreased her elbow extension after the intervention (Ch 12). In addition, in the follow-up assessment all children who received the TR training were able to maintain their improvements better than the children who received the NTR training. A previous well conducted RCT investigating the effect of task specific home based intervention using trunk restraint in adults with stroke (Michaelsen et al. 2006) reported an effect size of 1 for elbow extension after the intervention, with a minimal mean difference of 5 degrees extension for the group that received the TR. In contrast, in our study all children benefited from the intervention in terms of elbow extension. One reason why there was not a greater effect of TR on improvements in the range of elbow extension in our study, as has been reported for a similar intervention in adult stroke survivors might be that the underlying mechanisms of reaching and grasping impairments in children with CP are different from those after adult-onset brain lesions (Gramsbergen 2001; Hadders-Algra 2001). In adult-onset stroke it is possible that the combination of neurophysiological and biomechanical constraints that arise due to the lesions might prevent them from re-establishing well-coordinated arm and trunk movements during reaching that had been acquired previously. Thus, the application of trunk restraint may have permitted the system to re-experience previously learned movement coordination. On the other hand, considering the early brain lesions in children with CP and concomitant excessive plasticity where corticospinal tract connections may be replaced by extrapyramidal tracts in spinal segments, it is probable that children have not developed optimal movement coordination patterns. In adult stroke survivors and children with CP, excessive trunk involvement might serve a similar purpose with respect to assisting in the
transport of the hand to the target. The training outcome in children, however, may differ from that in adults since the children with CP are still learning or never acquired optimal UL coordination patterns during reaching tasks. Thus, directed practice in reaching with or without TR may have helped them to use elbow extension more effectively.

Trunk displacement

The main kinematic parameter that was affected by the intervention strategy was the amount of trunk displacement used while reaching to each target. Improvements were more marked in the children who received TR compared to NTR training. As well, for three individuals (Ch 2, 4 & 5) delayed improvement was observed at T2, as only at the follow-up evaluation was a marked improvement observed. By contrast, at the follow-up time period in children receiving NTR training, three children (Ch 7, 9 & 10) continued to deteriorate as did the child who improved post-intervention. In addition to the decrease in trunk displacement, four of the six children who received TR training also improved and maintained the improvement in elbow extension while this was not the case for the children who received NTR training. Indeed, while there was improvement in elbow extension for reaches to both targets in four of the six children in NTR, at the follow-up period, these gains were lost for most of the participants. Thus, the effect of task-oriented training with the trunk restrained not only resulted in better trunk use, but also on the use of the elbow during the follow-up period.

Does TR strategy lead to better UL movement quality?

Our results support the use of task-oriented training combined with trunk restraint to improve UL movement quality in children with CP. This is strongly supported by our findings that children who practiced movements with TR used less trunk displacement to compensate for arm deficits during reaching to close objects, and this behavior was maintained in the follow-up period. Furthermore,
children who participated in the TR training were able to maintain the improvements in hand trajectories and elbow extension in the follow-up assessment, while children who received NTR training lost the effect of the intervention or deteriorated at follow-up.

Some of the children who decreased trunk displacement in our study did not improve hand trajectory. This reinforces the notion that trajectory formation in children with CP may be the most difficult, or the last aspect of movement quality to improve. Perhaps longer training may result in more improvement in this parameter once the trunk displacement and elbow extension aspects achieve at a satisfactory performance level.

As well, some children who did not decrease trunk displacement increased elbow extension. These findings suggest that children could have used a different compensatory motor pattern than trunk forward displacement. There are several forms of motor compensations that should be considered during reach-to-grasp movement such as hand orientation, wrist flexion-extension, head position, trunk lateral inclination, and shoulder angles (Levin et al. 2004). We only addressed one of them in this study. With respect to compensatory movements, it should be noted that the most impressive result for the NTR group was the large effect size (range from 1.26 to 6.89) for the increase of trunk displacement post-intervention and three months following the intervention corresponding to a mean increase of 5 to 97 mm of trunk movement. Indeed, our results support previous findings (Schneiberg et al. 2004) that some children when engaged into UL practice increased trunk displacement as a form of compensation to improve reaching ability.

One advantage of using SSRD over group analysis is that we can address the specific question of which individuals can benefit the most from a specific form of therapy, instead of identifying a population effect. The disadvantage of SSRD is that it is difficult to estimate causality when more than one variable is investigated at the same time. More research is needed in order to determine the contribution of trunk movement to UL movement quality in children with CP and the relation between motor impairment and activity.
limitations. The obvious limitation of our study is the small sample size, which affects the generalizability of the findings. In order to increase the external validity of the results, this study needs to be repeated with a larger sample size. The study does however, provide some preliminary evidence as to the effectiveness of an UL intervention that includes restriction of excessive trunk movement. Our findings support the need to further investigate the effects of this type of intervention.

5.14. Conclusion

In this preliminary study, a task oriented intervention combined with trunk restraint, improved UL movement quality in children with CP. When the task oriented intervention was used with trunk restraint strategy, children decreased the amount of compensatory trunk displacement during reaching and grasping activities. However, when task oriented training was used alone (without TR strategy), the improvement in movement quality could be accompanied by an increase in compensatory trunk displacement.

5.15. Acknowledgments

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Wasiak J, Hoare B, Wallen M (2004) Botulinum toxin A as an adjunct to treatment in
the management of the upper limb in children with spastic cerebral palsy.
Cochrane Database Syst Rev: CD003469
Compensatory sprouting and impulse rerouting after unilateral pyramidal tract
lesion in neonatal rats. J Neurosci 20: 6561-6569
Table 4: Demographic and clinical characteristics of children with cerebral palsy task-oriented study.

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<thead>
<tr>
<th>Child</th>
<th>Sex</th>
<th>Age (years)</th>
<th>Impairment distribution</th>
<th>Most Affected UL</th>
<th>MACS</th>
<th>Semmes-Weinstein</th>
<th>2-points* (mm)</th>
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<th>Light Touch n / 20</th>
<th>Prop.‡ n / 8</th>
<th>CSI§ n / 16</th>
<th>PROM† n / 24</th>
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* 2-point discrimination, one child refused to make the test (na)
‡ Proprioception
§ CSI – Composite Spasticity Index
† Passive Range of Motion
Table 5: Number of points below the trend line indicating improvement of hand trajectory smoothness. Mean differences and effect sizes for smoothness of hand trajectories.
Children are grouped by the type of practice (TR=Trunk Restraint; NTR=No Trunk Restraint) and by target (T1, T2). The value indicated by the formula is the mean difference between intervention (\( \bar{x}_B \)) and baseline (\( \bar{x}_A1 \)), and the mean difference between the follow-up (\( \bar{x}_A2 \)) and the baseline (\( \bar{x}_A1 \)). Effect sizes (E.S.) are shown for each child by target and intervention assessment. Moderate to large effect sizes indicating improvement are in bold font and those indicating deterioration are shaded in grey.

<table>
<thead>
<tr>
<th>Child</th>
<th>TARGET 1</th>
<th>Post intervention (B)</th>
<th>Follow-up (A2)</th>
<th>TARGET 2</th>
<th>Post intervention (B)</th>
<th>Follow-up (A2)</th>
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</thead>
<tbody>
<tr>
<td></td>
<td></td>
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<td>E.S.</td>
<td>Points below baseline trend</td>
<td>E.S.</td>
<td>Points below baseline trend</td>
</tr>
<tr>
<td>TR</td>
<td></td>
<td>( \bar{x}_B - \bar{x}_A1 )</td>
<td>( \bar{x}_A2 - \bar{x}_A1 )</td>
<td></td>
<td>( \bar{x}_B - \bar{x}_A1 )</td>
<td>( \bar{x}_A2 - \bar{x}_A1 )</td>
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<td>0.15</td>
<td>9/10</td>
<td>-1.70</td>
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<td>0.78</td>
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<td>1.46</td>
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</table>
Table 6: Number of points above the trend line indicating improvement of elbow extension angle. Mean differences and effect sizes for elbow extension angle.
Children are grouped by the type of practice (TR=Trunk Restraint; NTR=No Trunk Restraint) and by target (T1, T2). The value indicated by the formula is the mean difference between intervention ($\bar{x}_B$) and baseline ($\bar{x}_A$), and the mean difference between the follow-up ($\bar{x}_{A2}$) and the baseline ($\bar{x}_A$). Effect sizes (E.S.) are shown for each child by target and intervention assessment. Moderate to large effect sizes indicating improvement are in bold font and those indicating deterioration are shaded in grey.

<table>
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<tr>
<th>Child</th>
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<th></th>
<th>TARGET 2</th>
<th></th>
</tr>
</thead>
<tbody>
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<td>Follow-up (A2)</td>
<td>Post intervention (B)</td>
<td>Follow-up (A2)</td>
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<td>Points above baseline trend ($\bar{x}_B - \bar{x}_A$)</td>
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Table 7: Number of points below the trend line indicating decrease in trunk displacement. Mean differences and effect sizes for trunk displacement.

Children are grouped by the type of practice (TR=Trunk Restraint; NTR=No Trunk Restraint) and by target (T1, T2). The value indicated by the formula is the mean difference between intervention ($\bar{x}_B$) and baseline ($\bar{x}_{A1}$), and the mean difference between the follow-up ($\bar{x}_{A2}$) and the baseline ($\bar{x}_{A1}$). Effect sizes (E.S.) are shown for each child by target and intervention assessment. Moderate to large effect sizes indicating improvement are in bold font and those indicating deterioration are shaded in grey.

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<th>Child</th>
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<th>Follow-up (A2)</th>
<th>Post intervention (B)</th>
<th>Follow-up (A2)</th>
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<tr>
<td></td>
<td>TARGET 1</td>
<td>TARGET 2</td>
<td></td>
<td></td>
</tr>
<tr>
<td>TR</td>
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<td>Points below baseline trend</td>
<td>Points below baseline trend</td>
<td>Points below baseline trend</td>
</tr>
<tr>
<td></td>
<td>($\bar{x}<em>B - \bar{x}</em>{A1}$)</td>
<td>($\bar{x}<em>{A2} - \bar{x}</em>{A1}$)</td>
<td>($\bar{x}<em>B - \bar{x}</em>{A1}$)</td>
<td>($\bar{x}<em>{A2} - \bar{x}</em>{A1}$)</td>
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Table 8: Numbers and percentage of children who improved kinematic variables and clinical scores following task-oriented with (TR) or without (NTR) trunk restraint.

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<th>Type of task-oriented training</th>
<th># Peaks</th>
<th>Elbow angle</th>
<th>Trunk</th>
<th>Melbourne (Target 2)</th>
<th>Total %</th>
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</tr>
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<td>Post interv.</td>
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<td>T1 T2 T1 T2</td>
<td>T1 T2</td>
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<td>1 1</td>
<td>72% 42% 67% 50%</td>
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<td>0 1</td>
<td>39% 29% 33% 25%</td>
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# Peaks: number of movement units in hand tangential velocity profile
Fig. 9: Schematic figure illustrating the wooden box used for the task-oriented training.

A) *Horizontal plane*; Zones to perform reaches to the close ipsilateral (I) and contralateral (II) side and to the far ipsilateral (III) and contralateral (IV) side of the most affected arm. B) *Vertical plane*, arrow marks shelf position at which reaches are done above 90° of shoulder flexion.
Fig. 10: Task specific block with virtual reality.
A) Demonstration of hand capture by the webcam. B) Birds and Balls application controls movement force and uses light touch in order to transform the ball into a bird; smashing the ball makes it disappear; B) Volleyball is played against the robot using a variety of upper limb movements. C) Soccer can be played with many hand positions; the aim is to defend the goal. The application can be progressed in difficulty.
Fig. 11: Trunk restraint

A) Side view of trunk restraint system. Shoulder straps pass through a hole behind the chair that was adjusted to the height of the spine of the child’s scapula. The box used for the task-oriented intervention and some toys are seen. B) Trunk restraint front view. Seat belt toys were put around the Velcro band to increase the child’s interest.

Trunk Restraint (TR)
Velcro Bands
Fig. 12: All data points observed in each child for reaches to Target 1 in each assessment. Baseline (A1), post intervention (B), and follow-up (A2). A regression line was drawn through all baseline points and extended through B and A2 assessments. Separate columns are used for the three kinematic outcome measures. A) Kinematic data are shown for (A) children 1 to 6 who received the task-oriented intervention with trunk restraint (TR) and (B) children 7 to 12 who received the task-oriented intervention with no trunk restraint (NTR).
Fig. 13: All data points observed in each child for reaches to Target 2 in each assessment. Conventions as in Fig. 4. A) Task-oriented training with trunk restraint (TR) B) Task-oriented training without trunk restraint (NTR).

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Fig. 14: Effect sizes for all children for the three kinematic parameters

Effect sizes for all children for the three kinematic parameters at the post-intervention assessment for hand trajectory smoothness (# peaks, open circles), elbow extension (triangles) and trunk displacement (filled circles). Children 1 to 6 received task-oriented training with trunk restraint (TR) and children 7 to 12 received task-oriented training without trunk restraint (NTR). Points below the effect size of zero (horizontal dashed line) represent improvement for trajectory smoothness and trunk displacement (negative effect size). Points above the dashed line (positive effect size) represent improvement for elbow extension. A) reaches to Target 1, B) reaches to Target 2.

**Summary Effect Size (E.S.)**
Fig. 15: Melbourne assessment scores for all children.
Melbourne assessment scores for all children at pre-test 1, pre-test 2, pre-test 3, post-test and follow-up assessment sessions. Horizontal dashed lines indicate the mean ± 2 SDs of the three pre-test Melbourne scores. A) Melbourne assessment scores for (A) children 1 to 6 who received the task-oriented intervention with trunk restraint (TR) and (B) children 7 to 12 who received the task-oriented intervention with no trunk restraint (NTR).
CHAPTER 6

DISCUSSION

Do rehabilitation strategies improve UL movement quality in children with CP? In order to answer this question, these study addressed the effect of two rehabilitation strategies on UL movement quality in children with CP: the use of arm constraint and trunk restraint. This study also determined the reliability of kinematic variables in a functional reaching task while aiming to use the most reliable measurement to investigate the effect of trunk restraint during task-oriented intervention. In this chapter, the findings of the three projects, which composed this PhD thesis, will be summarized, and then the significance and impact of the findings to scientific knowledge in pediatric rehabilitation will be explored, as well as their clinical applications. Finally, the limitations of the studies and suggestions for future research will be discussed.

6.1. Summary of findings

In the first study (manuscript #1), we aimed to determine whether improvements in clinical arm motor impairment following mCIT could be attributed to changes in arm movement quality as determined by detailed kinematic analysis. In addition, we hypothesized that some of the changes would be detected by kinematic analysis but not by the clinical scale such as QUEST. The main findings after mCIT were as follows: two of the three children improved their scores on the QUEST scale, and this improvement was accompanied by a significant change in one of the kinematic variables, trunk displacement. The child whose scores did not improve on the QUEST scale had an improvement in elbow extension after mCIT detected by the kinematic analysis. The most striking result was that the two children who had improved on the QUEST scale also increased their trunk displacement after mCIT. Overall, the results suggested that mCIT might improve some aspects of UL movement
quality. However, not all changes in movement quality were detected by a clinical QUEST scale. Indeed, the fact that two children improved motor function on QUEST, while increasing their trunk displacement as detected by the kinematic analysis, suggests that there is a possibility that constraint therapies improve activity via a corresponding increase in motor compensations. The fact that the third child improved elbow extension but did not improve on the QUEST further highlights the fact that this clinical outcome measure may not be sensitive to changes in movement quality implicit in the task of reaching. One other reason might be that the QUEST analyses movement quality statically, instead of dynamically. Therefore, kinematics analysis is more efficient to investigate movement quality, and more sensitive to motor compensations.

The second study investigated the test-retest reliability of kinematic variables of a functional reaching task. Three areas of the arm workspace were chosen in order to investigate the effect of target distance on kinematic variables. The test-retest reliability was measured via intraclass coefficients (ICCs). Results indicated that almost all kinematic variables evaluated had good reliability, with ICCs above 0.75. As expected, the reliability coefficients varied according to the workspace area or target distance where the reaches occurred. Shoulder angles had moderate ICC values because of the small ranges required in the reaching functional task. As only variables with ICCs above or equal to 0.90 should be selected to be used in clinical trials, three variables were retained for use in the functional reach study: hand trajectory smoothness, elbow extension, and trunk displacement. However, it should be noted that those variables can be used only if the kinematic task investigated is similar to the functional reaching task used in this study.

The purpose of the third manuscript was to investigate the effect of task-oriented intervention on UL movement quality, measured through kinematic analysis and the Melbourne clinical assessment. Our specific objective was to examine if a task-oriented intervention with a trunk restraint strategy was more effective than task-oriented intervention alone. Our first hypothesis was that task oriented training would improve UL movement quality in children with CP, and
that improvements would be maintained at three months following the end of the intervention. Our second hypothesis was that a better effect on movement quality with a decrease in movement compensations would be detected in children who received task-oriented training with trunk restraint strategy as compared to those who received task-oriented training alone.

The results suggested that task-oriented intervention with TR promoted better improvements in UL movement quality in children with CP than task-oriented intervention without TR strategy. In addition, some of the changes captured by the kinematic analysis were not in agreement with the Melbourne clinical scale. Although almost all children increased their elbow extension, only the children who received the task-oriented intervention with TR strategy were able to carry over their improvement three months after the intervention. Furthermore, some children who received the task-oriented intervention without TR doubled their pre-intervention trunk displacement levels.

6.2. Measuring movement quality in rehabilitation

An outcome measurement can be classified according to its goal. It can be evaluative, when the goal is to measure change. When the goal is to discriminate between groups or subgroups, the outcomes are classified as discriminative. When used to predict an individual’s future status or otherwise prognosticate, the outcome is predictive (Law and MacDermid 2008). In this study, our interest was in outcomes capable of measuring change after a treatment. Outcome measurements used to test change in this study were clinical outcomes (QUEST and Melbourne) and kinematic variable outcomes. All of these are outcome measurements that assess motor performance at a body function level according to the ICF model (Majnemer 2006). The Melbourne clinical assessment, and kinematic variables, measure performance in specific motor tasks and might be mistaken for outcomes that measure activity, when, in fact, they measure function or impairments. The QUEST scale measures quality of movement, but only in a static manner for the arm, without the context of activity. It is more dynamic in
measuring movement quality for the hand, within an active context (e.g. drawing). The impact of impairments on activity and participation has also been debated (Damiano 2008; Shikako-Thomas et al. 2008; Wright et al. 2008). Most of the authors agree that the relationship among different levels of the ICF are complex. Thus a direct relationship between changes in impairment and activity has not yet been proven. Furthermore, children’s personal factors and environmental context also play a role in activity and participation (Wright et al. 2008).

Despite the updated concept of the International Classification of Functioning, Disability and Health (ICF) (Rosenbaum and Stewart 2004), and the replacement of some terminology used in the International Classification of Impairments, Disabilities and Handicaps (ICIDH) (WHO 1980), there is still some confusion regarding some definitions of activity and function. Palisano et al. (2006) have defined the old model of classification as a disablement model, and they compared the terminology used in the two models (ICIDH and ICF). In the new model - the ICF - the term function is related to body, or physiological, functions, while activity refers to the performance of the task or action. Thus, activity consists of the integration of body functions. Unfortunately, there is still some confusion in the literature; some authors are still using the term function when they really mean activity (Damiano 2006; 2008, and many others). The source of confusion could be the old terminology used in the ICIDH, which at the personal level described functional limitations as the inability to perform some actions or tasks (WHO, 1980). Additionally, the word function has traditionally been used by therapists in rehabilitation to refer to the ability or capacity to perform a task. However, regardless of the ambiguity in the terminology, and in order to avoid further discussion about which definition of function or activity should be used, the definitions will hereby be interpreted according to context. According to Gordon (2007) the most important current concern in rehabilitation is how to measure activity. However, the question that appeared with the results of this study is whether measuring activity is sufficient to detect change due to a treatment. In addition, we must also inquire as to which clinical measure is responsive to a meaningful clinical change.
Gordon’s (2007) concern was related to the fact that most of the pediatric outcomes which aimed to measure hand function did not focus on activities. He also emphasized that the majority of outcomes are not designed to measure a unilateral impairment as in the case of hemiplegia. However, there are some well known pediatric outcome instruments that measure UL activity (Majnemer 2006). One of them is the Pediatric Evaluation of Disability Inventory (PEDI) (Haley et al. 1992). The PEDI measures activity in 3 domains: self-care, mobility, and socialization. Other examples of outcomes that measure UL activity are some parts of the Functional measure of independence for children (WeeFIM) (Bagley et al. 2007), the Canadian Occupational Performance Measure (COPM) (Law and Canadian Association of Occupational Therapists. 1998), and some parts of the School Function Assessment (SFA) (Davies et al. 2004). Although there are quite a few clinical evaluations of activity, those scales measure bimanual activities without deeper reference to the most affected arm. In fact, children with mild or even moderate hemiplegia score high on those scales. This argument is supported by the scores obtained with the PEDI scale in children with hemiplegia and diplegia that participated in this study and scored within the TD range (no reported results). The reason for the high score is that those scales do not account for the use of the most affected arm, nor for the use of motor compensations.

Gordon (2007) recently suggested a new scale: the Assisting Hand Assessment (AHA). The AHA was developed by a group at the Karolinska Institutet, Stockholm, Sweden (Krumlinde-Sundholm et al. 2007). The AHA measures performance, not capacity, because children are asked just to perform a specific bimanual activity. They are not required to function at their best performance. The AHA is scored by observation through video tape of 22 items with a 4-point criterion referenced scale (Krumlinde-Sundholm et al. 2007). The highest rating in the AHA scale is 4, which means ‘effective’, and the lowest is 1, which means ‘does not do’. The AHA scale has inter-intra rater (judge) reliability and can indicate differences in children’s abilities when used in tests with children with hemiplegic CP and obstetric brachial plexus palsy (Krumlinde-
The authors reported that the scale is still being investigated to prove validity, but the preliminary results are promising.

The possibility of outcome to measure activity shown by AHA is promising, but still does not answer all the problems in pediatric UL outcomes. In rehabilitation we require an outcome that measures activity, but it should also account for movement compensation. Moreover, we require an outcome tool that is able to detect meaningful clinical changes (responsiveness). Our results support the use of kinematic analysis to evaluate performance of an activity and to describe underlying impairments. In addition, our results demonstrated that some changes are captured by kinematic analysis but not by clinical outcomes. Finally, we also demonstrated in our two studies that improvements in clinical outcomes could be followed by motor compensations. The use of kinematic analysis in clinical settings is not feasible. It is costly and requires high level training. However, future clinical outcomes which evaluate function or activity should incorporate some elements of ‘how’ the movement was performed based in studies using kinematic analysis. Subjective-observational outcomes should be developed in a manner which maximally avoids the personal or rater judgment of the task: the more reliable the tool, the more sensitive to change the tool will be (Streiner and Norman 2006). However, a sensitive outcome is slightly different than a responsive one. Sensitivity to change is the ability of the tool to measure any degree of change, whereas responsiveness is the ability of a tool to measure clinically important change (Streiner and Norman 2003). The interest for rehabilitation here is to have outcomes that measure function within an activity context, include elements of ‘how’ activity is achieved, and have a reported responsiveness (Majnemer and Limperopoulos 2002).

6.3. Early brain lesions and Motor compensations

Children with CP, who have large lesions involving the cortex and subcortical white matter, often have important impairment in UL (Kulak et al. 2006). The occurrence of UL motor and sensory impairments in early ages can
have harmful consequences to development (Johansson and Cole 1992; Cooper et al. 1995; Gordon and Duff 1999). For example, these impairments may result in a learned nonuse phenomenon (Taub et al. 1994; Taub et al. 2006). The presence of ‘excessive’ plasticity in early stages of development might also induce formation of abnormal motor behaviors (Johnston 2004).

Children with UL impairment that limit activity tend not to use their affected limb (Charles and Gordon 2005). Unsuccessful attempts to use the most impaired limb can lead to a ‘developmental disuse’, a learned nonuse acquired during development. The phenomenon of learned nonuse was first observed by Taub (1977) from neurophysiological and behavioral studies with deafferented monkeys. When the sensation was eliminated in one of the monkey limbs, this limb was never used again in a free situation.

After the use of a restraint in the unaffected limb or during a training situation the monkey returned to use his affected limb (Taub et al. 2006). The authors explained that one possible reason could be motivation. According to them, the conditioning of restraint obliges the monkeys to use the affected limb, or in the case where the monkey did not use the affected limb, it would be hungry or would receive an electrical shock. The other suggested explanation was to account for a theoretical existence of an inhibitory mechanism between the two limbs, where the movements of the intact limb could have an inhibitory effect on the movements of the deafferented limb. The suggested inhibitory mechanism would be only present in abnormal physiological situations, such as when one of the limbs had a disruption in its afferent information. This theory has not been proven in an empirical study.

Another study involving monkeys that were deafferented during perinatal stages and postnatal stages also has reported benefits of the use of restraints in the less affected limb (Taub et al. 1975). The arm restraint strategy, born from the learned nonuse hypothesis, evolved and became the constraint-induced movement therapy (CIT) (Taub et al. 1999). In children with CP, all studies done to date have reported benefits from CIT or a modified form of CIT (Charles and Gordon 2005), but the confidence in the evidence is still moderate to low. Charles and
Gordon (2005) found 15 clinical trials, none of those was classified as an RCT with a narrow confidence interval, or level 1b of evidence (Law and MacDermid 2008). Furthermore, all of them used outcome measurements that did not account for motor compensations (Charles and Gordon 2005).

In the first study presented in this thesis (the three case reports) we investigated the effect of mCIT on UL movement quality. Two of the children who improved on the QUEST scale increased their trunk displacement. There is much neuroanatomical and neurophysiological evidence of the high plasticity during infancy and childhood (Huttenlocher 1979; Huttenlocher 1984; Chugani et al. 1987; Chugani 1998). This evidence, combined with the fact that children with CP never experienced how to use their affected arm as do TD children, is supported by findings demonstrating that there is a difference in sensory and motor cortical organization following brain lesions in early life (Kulak et al. 2006). This further raises the question of whether or not the phenomenon of learned nonuse in children with hemiplegic CP have the same mechanisms as found in adults with hemiplegia, even considering that learned nonuse in children might be called ‘developmental disuse’ (Charles and Gordon 2005). It has been reported that in children with CP there is an inter-hemispheric difference in the organization of sensory and motor pathways, suggesting that motor impairments in the affected UL could be due to dissociation between the normal sensory and motor representation in the cerebral cortex, following neural organizations after early brain injury (Maegaki et al. 1999; Thickbroom et al. 2001). In other words, children with CP might have deficiencies in associating kinesthetic inputs with motor outputs. Therefore, a restraint in the less affected arm should be employed with reservation in this population, so as not to trigger motor compensations. In addition, three of the children in the task-oriented study have deteriorated performance in hand trajectories after intervention, and all three children had lower scores in the sense of position and other sensory tests. This finding suggests that in children with sensory deficits hand trajectory improvement strategies must be linked with other strategies facilitating integration of sensory and motor inputs during motor performance.
6.4. Motor compensations in children can be more than just maladaptive behaviour

Motor compensations in adults have been related to neurophysiological and biomechanical deficits (Levin et al. 2002). In children with CP, there are some muscle and skeletal changes during growth that can lead to deformities by the presence of maladaptive behaviors (Hof 2001; van Eck et al. 2008), especially due to hypertonia. However, no relationship between the growth of the skeletal system during puberty and gross motor function deterioration was found (van Eck et al. 2008). Motor compensations in children might have other origins than just maladaptive behaviors.

The dynamic systems theory applied to motor development (Thelen 1995; Smith and Thelen 2003) embraces the theory of neuronal group selection (NGS) (Sporns and Edelman, 1993). The NGS theory, as in traditional Darwinian theory, proposes that a selectionist view of development requires a source of diversity and variability from which adaptive patterns can be chosen. The NGS further proposes that the development of sensorimotor coordination occurs in three steps: 1. Spontaneous generation during development of a variety of movements forming a basic movement repertoire; 2. development of the ability to sense the effects of movements in the environment, subsequently guiding neural selection; and 3. the actual selection of movements. Selection in the nervous system is mediated by synaptic change, resulting in the stabilization of brain circuits that support the specific goal-directed movements. The somatic selection in the nervous system results from the competitive strengthening of neural connections (synapses) involved in the generation of successful movements (Sporns and Edelman, 1993). Children with CP, when discussed using NGS theory (Hadders-Algra 2001), may have problems selecting the most efficient neural repertoire to control the execution of voluntary movements. However, they are still developing, and probably, in spite of their kinesthetic deficits, they can still take advantage of the ‘redundancy’ of their systems (Bernstein 1967, Latash and Turvey 1996) and use degrees of freedom that are not used by TD children when they move. According to Thelen (1995), the systems theory (Bernstein 1967)
proposes that the subsystems and components that produce movement are assembled from whatever segments and joints are available to fit the task. This organisation gives the system great flexibility while meeting the demands of the task within a continually changing environment, all while keeping the goal of the movement in mind.

Feldman (1998) has proposed that the nervous system generates movements by combining independent efferent control inputs with afferent inputs from vestibular, visual, and proprioceptive receptors. This forms the actual frame of reference (FR) of the body and its surrounding external space. The FR changes its orientation (voluntary movement) when the values that constitute the original FR change (efferent and afferent inputs). The author gave an example that a toddler’s ability to stand can only be acquired when the ability to form a united FR by integrating all intrinsic (body) and extrinsic (space) information is completed. Feldman’s (1998) theory of FR is similar to Schmidt’s theory of motor schema (1975). Schmidt’s schema theory state that motor learning is improved with varied practice, which in his concept would generate more diversified motor programs. Both FR and Schema theory states that a movement skill is acquired though accumulating sensory and motor experience. The literature available in motor development (Thelen 1995) and the abnormal neurophysiological findings (Kulak et al. 2006) thus support the notion that motor compensations in children with CP are not simply just maladaptive behaviors, but that they can also be the result of a maladaptive plasticity. One way to overcome this issue is to develop treatments where a varied motor practice regime includes the prevention of motor compensations. Based on this rationale, this study investigated the effect of task-oriented intervention with trunk restraint on UL movement quality.

6.5. Is the trunk displacement during reaching in children with CP compensatory?

It was observed that adults who had sustained a stroke and subsequently had limitations in shoulder and elbow range of motion could perform reach-to-
grasp tasks in an accurate manner. Those tasks were only accomplished because the transport of the hand to the target was mostly carried out by excessive trunk motion (Cirstea and Levin 2000; Michaelsen et al. 2001). In children however, in addition to the possibility of compensation for motor impairments and biomechanical constraints, excessive trunk displacement might be explained by the fact that during development the nervous system did not experience the sensory information necessary to build a “postural schema” observed in typically-developing children where the arm segment becomes more efficiently involved in the task and the excessive trunk displacement decreases with age (Schmidt 1976; Schmidt et al. 1992; Feldman and Levin 1995; Schneiberg et al. 2002; Schmidt 2003). This theory has its basis in the findings that children with CP have impaired sensory motor integration (Maegaki et al. 1999; Thickbroom et al. 2001).

Little is known about the development of reaching in children with CP. Several studies characterized reaching in children with CP at points in time, but not longitudinally (Van Thiel and Steenbergen 2001; van Roon et al. 2004; van der Heide et al. 2005), and none of them investigated arm and trunk coordination during reaching. The excessive trunk displacement observed in children with CP is considered compensatory because of findings from previous studies where children with CP performed reaching and grasping activities and their UL movements occurred together with excessive trunk movements (Steenbergen et al. 2000; Volman et al. 2002; van Roon et al. 2004; van Roon et al. 2005; Mackey et al. 2006; Coluccini et al. 2007; Kreulen et al. 2007); the amount of trunk movement observed in children with CP was twice the amount of trunk movement observed in normally developing children with the same age (van Roon et al. 2004, 2005, Mackey et al. 2006). In addition, excessive trunk displacement is considered compensatory in children because its elimination is associated with better UL function (van Roon et al. 2005; Kreulen et al. 2006; Kreulen et al. 2007). Furthermore, the results of two studies demonstrated in this doctoral thesis suggest that UL rehabilitation intervention that did not prevent trunk compensations might in fact increase them. Therefore, excessive trunk
displacement should be considered compensatory to UL impairments in children with CP and must be prevented in rehabilitation interventions aimed at improving UL function.

6.6. Evidence based on single subject research design

The literature review in this study has described the heterogeneity of children with CP in many aspects from etiology to clinical symptoms. Still, variability is present even in the standardized classification measurement scales MACS and GMFCS (Tieman et al. 2007; Arner et al. 2008). Although these measurements have contributed enormously to the facilitation of communication in research and clinical practice (Steenbergen 2006; Gunel et al. 2008), stratifying children with CP in clinical trials by their functional ability classification does not decrease the variability of their impairments or activity problems. In addition, it has been suggested that outcomes and treatments should account for the age-skill acquisition process (Majnemer and Limperopoulos 2002; Majnemer and Mazer 2004), and that development in children with CP has been reported to be associated by the type of impairment distribution (Hanna et al. 2003; Arner et al. 2008). As children with CP are such a heterogeneous population, why is there still a tendency to generalize the scientific evidence and treatment related to this population? Moreover, how do we demonstrate the effect of treatment with group effects, if this population does not react to treatment in an average (and thus predictable) fashion? The presence of large differences in individuals’ changes and responses to treatment influences the validity of the demonstration of a group treatment effect (Streiner and Norman 2006). In other words, carrying out a group comparison approach in a heterogeneous group could mistakenly attribute the effective kind of treatment to an extreme response or outliers. According to Dr. Nancy Mayo (in a personal communication) when the sample size is not large and the group is highly heterogeneous, not only will the group approach not have enough power to detect even large effects, but also the extreme responses in either of the groups can negate any effect.
Because of the high heterogeneity of children with CP, the use of single subject research design (SSRD), or randomized n of 1 trial, combined with highly sensitive outcome measurements is preferable to group comparisons for investigating treatment effects. Furthermore, SSRD, when carefully conducted by respecting internal validity issues, can be considered an optimal design to implement scientific evidence to practice (Patrick et al. 2000). The increasing need to promote evidence based practice (Law and MacDermid 2008) demands a deeper evaluation of the criteria considered in determining evidence-based guidelines. Slavin (1995) has reported errors in the conclusions of some meta-analysis studies and has suggested that the evidence should be based on the methodology, characteristics of the study, validity of the results, content and procedures. According to Patrick et al. (2000) “while scientists are debating the standards of scientific evidence, people are suffering and needing services”. The authors have also added that the rate of knowledge transfer of scientific findings to clinical practice is, at the moment, poor.

Perhaps SSRD studies are not yet incorporated into the evidence based guidelines because of the lack of evaluative scales to critically review the methodology and validity of the results in the SSRD. Recently, however, two scales have been developed (Romeiser Logan et al. 2008; Tate et al. 2008). The Single-Case Experimental Design (SCED) scale is formed by 11 items (Tate et al. 2008). It has a content of validity comparable to the PEDro scale (The Physiotherapy Evidence-Based Database); the content validity was empirically tested in 85 published reports. Inter-rater as well as item reliability was reported to be excellent for individual raters, consensus rating, and between pairs of raters. Tate et al. (2008) have suggested that the SCED scale should be used as a checklist in critical appraisal and publications of SSRD. The other scale focuses on the level of evidence provided in the SSRD study (Romeiser Logan et al. 2008). However, the authors have also developed 14 questions to evaluate the quality of SSRD based on other critical appraisals of group designs. Each ‘yes’ in a question represents a single point; except for questions 5 and 8 were there are sub questions, where 0.5 is assigned for each sub item. Scoring cut-offs were
established: at 11 to 14 points the study was considered strong, 7 to 10 moderate, and less than 7 was considered weak.

The point that should be retained is that evidence based practice includes elements which are beyond the critical evaluation of research design; the evidence should be integrated within clinical judgments and with that most important of elements: the values of the patient and his/her individuality (Romeiser Logan et al. 2008). Furthermore, in order to promote best practice, it is necessary to address the demands of clinicians. Therefore, new designs should be developed where individual responses to treatment are addressed in a scientific way (Payton 1994).

“A great difficulty in developing the clinical science of physical therapy is that we treat individual persons, each of whom is made up of situations which are unique and, therefore, appear incompatible with the generalizations demanded of science.” This quote from Helen Hislop to the American Physical Therapy Association was reprinted in Domholdt’s book in the chapter for single-system designs pg.125 (Domholdt 2000).

6.7. Significance and clinical applications.

Considerable efforts to understand and treat upper limb function in children with CP have been made in the past years. At present however, important gaps remain in efforts to improve the efficacy of rehabilitation interventions in this population. The lack of outcomes describing movement performance and the inability to detect meaningful clinical changes is still a challenge to be overcome. In addition, outcome measurements that evaluate activity without assessing how movement is performed have the risk of being interpreted as successful task achievement and improvement of arm function, despite the fact that function in the arm improved but the task was achieved through movement compensation.

The results of this study have demonstrated that children with CP can perform tasks with the recruitment of excessive trunk movement, and that the clinical outcomes available which evaluate movement quality are not all able to
detect all of the details of movement performance. These findings increase the awareness of the importance to improve outcomes research for the pediatric population. More outcomes should be developed for the pediatric population, including developmental criteria, movement performance based in kinematic analysis studies and responsive to change.

Clearly, the results of this study also demonstrate how important it is to prevent motor compensations during upper limb rehabilitation training. The high plasticity present in early ages and the dynamic course of development make children adaptable to any intrinsic or extrinsic change. This adaptability can be harmful to skill acquisition and coordination if not guided in a proper manner. The positive effect obtained with task-oriented intervention combined with a trunk restraint strategy support the idea that clinicians should be aware of motor compensation when training arm and hand functions.

6.8. Limitation

An obvious limitation of this study was sample size. For the mCIT study, the sample of three children was convenient for verifying the feasibility of the CIT method with young children for future implementation in clinical setting. In the second and third study only 12 children were included because in the case of child-13, some important clinical measurements which are important elements for the SSRD were not collected.

Two factors might explain the small sample size of this study. First, none of the 5 pediatric centers have a database or registry of children with CP. Usually, recruitment was done by therapists who used recent files or waiting lists to select possible candidates. Second, in order to participate in the study the child had to stop and not be submitted to any other rehabilitation treatment targeting the upper limb. This element of inclusion is necessary to avoid contamination by other treatments, but at the same time was problematic. Children with CP who had severe limitations in UL activities were already engaged in a rehabilitation treatment for the UL. Thus, among children with UL deficits, we only could
recruit the ones who had already received treatment and still wanted more, or the
ones who were on a waiting list.

Another limitation of this study consisted of the number of data points
collected in the kinematic analysis. For the baseline assessment often we had
more than 15 points of data because the observations were made in three
assessments. However, for the post-test and follow-up we collected a minimum of
10 observations for each assessment, yet because of technical problems with the
data, some of the outcomes did not have the same number of observations. For
example, in the same assessment we recorded 12 trials for hand trajectories, and
only 7 trials for the elbow. This happened because markers used to determine
elbow angles were missing more frequently during the reaches while hand
markers were more visible for most of the trials. This made the interpolation
process difficult.

Finally, more information about the site of lesion, time of diagnosis,
assessments of activity, and subject participation would be optimal to add to the
clinical characteristics of the children. The PEDI self-care domain was used in
this study, but the results were not reported because most of the children had TD
scores. We used the scaled score in the PEDI, since the raw score can only be
used in children aged up to 7 years.

6.9. Future research

Research in CP is undoubtedly needed at different levels, from prevention
and early diagnosis, to activity and social participation. The following are the
most important to rehabilitation: 1. better outcomes assessing activity and
developed specifically for children with CP, taking in consideration the
impairment distribution (unilateral, bilateral), skill acquisition and developmental
rules, describing motor performance, and motor compensations; Determine the
sensitivity of UL kinematics in children with CP  2. better descriptions of the
development of children with CP, more longitudinal studies, and single subject
design all seem to be feasible approaches for this aim; 3. investigation of arm and
trunk coordination during reaching movements with experimental procedures
designed to test how the hand trajectory is stabilized if the trunk is arrested, as it
has been researched with adults (Adamovich et al. 2001; Ghafouri et al. 2002); 4.
more rehabilitation strategies and innovative treatments to promote improvement
in UL functions and activity should be developed and investigated.
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