Université de Montréal

Muscle co-activation during gait in children with cerebral palsy

par

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Ce mémoire intitulé

Muscle co-activation during gait in children with cerebral palsy

Présenté par

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A été évalué par un jury composé des personnes suivantes

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Résumé

La paralysie cérébrale (PC) est un trouble non progressif causé par une lésion cérébrale. La PC survient tôt dans la vie et présente une atteinte hétérogène et une altération fonctionnelle. Chez les personnes atteintes de PC, les modifications du Contrôle neuronal et des muscles entraînent des modifications permanentes de la fonction motrice, entraînant des déficits de mouvement. L'une des raisons des patrons de marche atypiques chez les enfants atteints de PC est l'altération l'activation musculaire. Un niveau anormal d'activation simultanée des muscles agonistes et antagonistes des muscles agonistes et antagonistes entourant une même articulation la même articulation empêche une performance de marche optimale chez les enfants atteints de PC. Ce phénomène est connu sous le nom de co-contraction musculaire (CoM) ou de co-activation musculaire (CaM) dans toutes les études. L'identification des schémas musculaires les plus détériorés, à savoir CoM/CaM, chez les enfants atteints de PC est essentielle pour une rééducation efficace de la marche. L'objectif de ce projet de maîtrise était donc de distinguer CoM/CaM chez les enfants atteints de PC de leurs pairs en développement typique (DT) pendant la marche. Cet objectif a été atteint en deux étapes ; Premièrement, nous avons décrit la CoM/CaM chez les personnes atteintes de PC via la réalisation d'une revue de littérature ; Ensuite, nous avons appliqué nos résultats de la première étape à une étude transversale pour comparer CoM/CaM pendant la marche entre des enfants atteints de CP et de DT.

Une revue de littérature suivant la méthodologie en 6 étapes du Joanna Briggs Institute a été effectué. Les bases de données ont été consultées à l'aide de mots-clés pertinents. Toutes les études publiées sur CoM/CaM chez les personnes atteintes de PC pendant la marche ont été recueillies. Après un examen de la pertinence des titres et des résumés, un deuxième examen des textes intégraux des sources par deux examinateurs a été appliqué. Enfin, les données ont été extraites des articles inclus (n=21). Ensuite, à l'étape suivante, les principales méthodes utilisées pour quantifier la MCa identifiées à l'étape précédente ont été codées dans Matlab (The Mathworks Inc., Natick, États-Unis) et appliquées à nos données de 12 enfants atteints de CP et 23 enfants TD. Nous avons comparé le CaM moyen de deux groupes de muscles de la cuisse et de

la jambe (Rectus Femoris (RF)/Semitendinosus (ST) et Tibialis Anterior (TA)/Lateral gastrocnemius (LG), respectivement) via des tests t non appariés (ou son équivalent non paramétrique).

La revue de littérature a suggéré une CaM plus élevée chez les personnes atteintes de PC par rapport à leurs pairs en bonne santé dans toutes les études. Bien qu'il y ait eu une terminologie et des approches méthodologiques incohérentes, nous avons pu discriminer les terminologies (c'est-à-dire CoM et CaM) en fonction des méthodologies de calcul (c'est-à-dire moment et EMG) utilisées par les études. En outre, cette étude nous a permis de résumer les modèles de CaM chez les individus atteints de PC et d'identifier la relation entre certains des paramètres de marche avec CaM. Enfin, les résultats de cette étude ont révélé des informations précieuses concernant les lacunes de la recherche dans ce domaine.

La deuxième étude a identifié une augmentation de la CaM pendant la marche (la foulée entière, la phase d'appuie et la phase oscillante) chez les enfants atteints de PC par rapport à leurs pairs TD. Cette augmentation n'a été observée que dans les muscles de la jambe (pendant la phase d'appuie et la phase oscillante) et dans les muscles de la cuisse (pendant la phase oscillante) lorsque nous avons normalisé les signaux d'électromyographie. Les groupes CP et DT n'avaient pas de CaM différent en utilisant l'EMG normalisé pour l'ensemble de la foulée. Cette différence met en évidence l'effet de la normalisation EMG sur les valeurs de CaM. De plus, les enfants avec le niveau II du système de classification de la fonction motrice globale (SCFMG) avaient un CaM plus élevé dans les muscles de la cuisse pendant le swing que ceux avec le niveau I.

Dans l'ensemble, ce projet de maîtrise révèle de nouvelles preuves soutenant une plus grande CaM chez les enfants atteints de PC par rapport à DT pendant la marche. Néanmoins, il est important d'étudier la CaM dans différentes phases de marche car elle affecte la comparaison entre les groupes. En outre, ce projet justifie l'importance de la méthodologie (par exemple, le traitement EMG et le calcul CaM) dans les études CaM. Plus précisément, il est fort probable que les résultats changent avec différentes approches de normalisation EMG. De plus, les enfants atteints de SCFMG I et II peuvent éprouver différents niveaux de CaM pendant la phase oscillante. Davantage de comparaisons dans des recherches futures, telles qu'entre les SCFMG I, II et III dans la PC hémiplégique et diplégique pendant les sous-phases de la marche (le contact initial, le « mid-

stance »), peuvent fournir de meilleures informations sur les modèles de CaM dans cette population.

Mots-clés : activité musculaire, analyse électromyographique, développement typique, marche, moment, normalisation, paralysie cérébrale, Revue de littérature

Abstract

Cerebral palsy (CP) is a nonprogressive disorder caused by a brain injury. CP occurs early in life, before, during, or after birth, and has heterogeneous involvement and functional impairment. In individuals with CP, changes in neural drive and muscles lead to lifelong changes in motor function, leading to movement deficits. One of the reasons for atypical gait patterns in children with CP is altered muscle activation patterns. An abnormal level of simultaneous activation of agonist and antagonist muscles crossing the same joint prevents optimal gait performance in children with CP. This phenomenon is known as muscle co-contraction (MCo) or muscle co-activation (MCa) across studies. Identification of the most deteriorated muscular patterns, namely, MCo/MCa, in children with CP is vital for effective gait rehabilitation. The objective of this master's project, therefore, was to distinguish MCo/MCa in children with CP from their typically developing (TD) peers during gait. This objective was achieved through two studies; first, we described MCo/MCa in individuals with CP via the conduction of a scoping review; then, we applied our findings to inform a cross-sectional study to compare MCo/MCa during gait between children with CP and TD.

A scoping review following the 6-stage Joanna Briggs Institute methodology was conducted. Databases were searched using relevant keywords. All published studies on MCo/MCa in individuals with CP during gait were collected. After title and abstract relevance screening, a second screening for the full texts of the sources by two reviewers was applied. Finally, data were extracted from the included articles (n=21). Then, leading methods used to quantify MCa identified from the previous study were coded in Matlab (The Mathworks Inc., Natick, USA) and applied to our data from 12 children with CP and 23 TD children. We compared the average MCa of two thigh and shank muscle groups Rectus Femoris (RF)/Semitendinosus (ST) and Tibialis Anterior (TA)/Lateral gastrocnemius (LG), respectively, via unpaired t-tests (or its non-parametric equivalent).

According to our scoping review, higher MCa in individuals with CP compared to healthy peers across studies was found. Although there were inconsistent terminology and methodological

approaches, we could discriminate terminologies (i.e., MCo and MCa) according to the methodologies in the calculation (i.e., moment and EMG) used by studies. Also, this study enabled us to summarize MCa patterns within individuals with CP and identify the effect of the some of the gait parameters on MCa. Finally, the findings of this study revealed valuable information regarding the research gaps in this area.

The second study identified increased MCa around the knee and ankle joints for the following muscles (i.e., RF/ST and TA/LG, respectively) during walking (i.e., entire stride, stance, and swing) in children with CP compared to their TD peers. This increase was seen only in shank muscles (i.e., during stance and swing) and in thigh muscles (i.e., during the swing) when we normalized electromyography (EMG) signals. CP and TD groups did not have different MCa using normalized EMG for the entire stride. This difference highlights the effect of EMG normalization on MCa values. Also, children with Gross Motor Function Classification System (GMFCS) level II had higher MCa around the knee during swing than those with level I.

Overall, this master's project reveals new evidence supporting greater MCa in children with CP compared to TD peers during walking. Nevertheless, it is recommended to investigate MCa within different gait phases as it affects the comparison across groups. Also, this project justifies the importance of methodology (e.g., EMG processing and MCa calculation) in MCa studies. More specifically, it is likely that the results alter with different EMG normalization approaches. Moreover, children with GMFCS I and II can experience various levels of MCa during the swing phase. More comparisons in future research, such as between GMFCS I, II, and III in hemiplegic and diplegic CP during gait sub-phases (i.e., initial stance, mid-stance), can provide better information regarding MCa patterns in this population.

Keywords: Cerebral palsy, Electromyography analysis, Normalization, Scoping review, Moment, Muscle activity, Typically developing, Walking

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List of acronyms and abbreviations

CP: Cerebral palsy

MCo: Muscle co-contraction

MCa: Muscle co-activation

TD: Typically developing

RF: Rectus Femoris

ST: Semitendinosus

TA: Tibialis Anterior

LG: Lateral Gastrocnemius

EMG: Electromyography

GMFCS: Gross Motor Function Classification System

CGA: Clinical gait analysis

So: Soleus

SENIAM: Surface Electromyography for the Non-Invasive Assessment Muscles

RMS: Root Mean Square

MVC: Maximum voluntary contraction

VM: Vastus Medialis

VL: Vastus Lateralis

GM: Gastrocnemius Medialis

BF: Biceps Femoris

MH: Medial Hamstring

EEI: Energy index expenditure

To my parents and my husband, for their endless love

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This dissertation is only the beginning of another journey!

Chapter 1 – [Introduction]

Cerebral palsy (CP) occurs every three to four per 1000 live births in the United States (Yeargin-Allsopp et al. 2008). Individuals with CP have lifelong motor impairments and disabilities (Aisen et al. 2011). There are different classifications for individuals with CP according to the type of disorder (e.g., spastic, ataxic) and the involved body segment (e.g., hemiplegia, diplegia, quadriplegia) (Minear 1956). One of the most important consequences of CP is an abnormal gait pattern, and patients with CP may exhibit a wide range of gait deviations. Understanding the relationship between gait deviations and impairments in the population with CP would help gait rehabilitation (Armand, Decoulon, and Bonnefoy-Mazure 2016).

The contraction of different muscle groups drives walking, and muscular adaptations are needed to enable humans to walk on different surfaces at the required speed (David A. Winter 2009a). In order to understand the action of the agonists or antagonists during a given activity, we can study electromyography (EMG) profiles (David A. Winter 2009a). During some movements, agonist and antagonist muscles crossing the same joint activate simultaneously, which is known as muscle co-activation (MCa) (Diane L. Damiano 1993) or muscle co-contraction (MCo). In normal movement, a certain amount of MCo/MCa helps stabilize the joint, for example, when lifting weights or walking or running (David A. Winter 2009a). However, sometimes it could be problematic; for example, MCo/MCa occurs, potentially more frequently and with greater intensity, in many pathologies, such as stroke patients (Lamontagne, Richards, and Malouin 2000), and particularly in individuals with spastic CP (David A. Winter 2009a).

Individuals with CP suffer from a lack of selectivity, reduced coordination and balance, and muscle weakness (Bax et al. 2005). It has been shown that altered muscle size and co-activation patterns lead to muscle weakness in individuals with CP (Hussain et al. 2014). That is, abnormal MCo/MCa has detrimental effects on the functionality of children with CP during gait (Leonard, Hirschfeld, and Forssberg 1991; Unnithan, Dowling, Frost, Volpe Ayub, et al. 1996). Although gait analysis and EMG patterns have been studied extensively in children with CP, relatively little attention has been paid to the contribution of different MCo/MCa levels seen during walking in this population.

Moreover, studies have been inconsistent in quantifying and interpreting MCo/MCa. Thus, quantifying MCo/MCa in children with CP and comparing it with typically developing (TD) children is needed, and it might improve clinical gait management in this population.

Understanding the impact of MCo/MCa on functional ability may help to develop more appropriate therapeutic interventions in children with CP. Treatment plans might be affected by the heterogeneity of the abnormalities in children with CP. For instance, it is argued that MCo/MCa should only be treated if it is a tertiary abnormality (i.e., compensation) and not a primary abnormality (i.e., impairment) (Gage et al. 2009). That means understanding the difference between MCo/MCa patterns during gait in children with CP and TD peers and the role it plays in the walking skill of these children could assist decision-making in rehabilitation. Therefore, the objective of this master's project is to (1) describe, via a scoping review, MCo/MCa during gait in individuals with CP and (2) apply these findings to compare MCo/MCa during walking in children with CP with their TD peers in a cross-sectional observational study. The Hypotheses are (1) included studies will describe MCo/MCa during gait in individuals with CP (non-directional) and (2) MCo/MCa will be increased in children with CP compared to their TD peers.

Structure of the master's project

This project is presented in six chapters. This first introductory chapter is followed by a literature review (Chapter 2) describing the main concepts relevant to the study of MCo/MCa in individuals with CP. In the third chapter, MCo/MCa during gait in individuals with CP is presented in the form of a scoping review (submitted to Gait and Posture, October 2022) following the 6-stage Joanna Briggs Institute methodology (Peters et al. 2017). In the fourth chapter, leading methods used to quantify MCo/MCa identified from chapter three were coded in Matlab (The Mathworks Inc., Natick, USA) and applied to a dataset from 12 children with CP and 23 TD peers. The fifth chapter presents a general discussion on the perspectives offered by this master’s project. Finally, the sixth chapter is devoted to the overall conclusions.

Chapter 2 – [literature review]

This chapter is divided into four sections. It starts with a description of gait analysis which will allow a general understanding of the biomechanical characteristics of gait, primarily muscular activation occurring during this movement (section 2.1). Then, the following section describes in-depth the muscle activity measurement considerations (section 2.2). Furthermore, this section focuses on two concepts of muscle co-activation (MCa) and muscle co-contraction (MCo), considering the current literature. In the next section, we provide background on Cerebral Palsy (CP), followed by a description of muscular pathology and gait patterns in this population (section 2.3). In this section, the gait patterns in individuals with CP are explained to understand better the gait adaptations resulting from the impairment. Finally, this chapter concludes by stating the main objectives of this master project (section 2.4).

2.1 Gait analysis

2.1.1 Background

One of the main activities in human locomotion is walking (Chiou and Burnett 1985), which aims to displace the whole body between two places (Perry 1992). Muscular activation and joint movements contribute to optimized motor control and smooth walking, and any disruption in human function, for example, illness or pain, brings about restricted movement patterns (Jessica Rose and Gamble 2006).

Over the last decades, gait analysis has been used to evaluate the functionality of walking. Gait analysis is defined as the quantitative study of all mechanical features of walking using laboratory facilities and systems (Cappozzo 1984). Visual observation, quantitative measurement, and biomechanical analysis are three main disciplines that have been suggested by modern gait analysis (Saleh and Murdoch 1985), and until 1992, there were five measurement systems for gait analysis as Motion analysis, electromyography, ground reaction forces, stride analysis, and energy cost measurements (Perry 1992). Today, advanced laboratory gait assessment is conducted using motion capture, reflective markers, and force plates to evaluate normal and pathological gait.

Researchers use cutting-edge technology and software to represent and interpret gait, advancing human knowledge about this vital task. A combination of observational and instrumented analysis contributes to a more in-depth gait assessment, particularly in helping quantify gait deviations (Jessica Rose and Gamble 2006).

Some people suffer from gait abnormalities. Thanks to the advanced technology used in Clinical gait analysis (CGA) in the last two decades, it has been possible to conduct objective, accurate, and more sensitive disease assessments (Shanahan et al. 2018). In CGA, the goal is to quantify the characteristics of pathological gait (Wren et al. 2011) using information from four primary sources of data: spatiotemporal, kinematics, kinetics, and electromyography (EMG) (Armand, Decoulon, and Bonnefoy-Mazure 2016). Besides recognizing gait deviations, CGA plays a key role in studying the impact of various treatment modalities on gait disorders. For example, gait management in children with CP has improved with the knowledge from CGA (Armand, Decoulon, and Bonnefoy-Mazure 2016).

2.1.2 Gait phases

A gait cycle is a sequence of movements in which the body weight is transferred from one limb to another to reach the destination. During a gait cycle, one leg helps the advancement of the body while the other supports the body weight (Perry 1992). The gait cycle is also known as a “stride” (Murray, Drought, and Kory 1964). As Perry (1992) described, stride length is the distance between the first and second contact by the same foot. Another term, “step,” has been introduced to describe the length of the distance between the initial contact of each foot (i.e., right and left) (Perry 1992) (Figure 1)

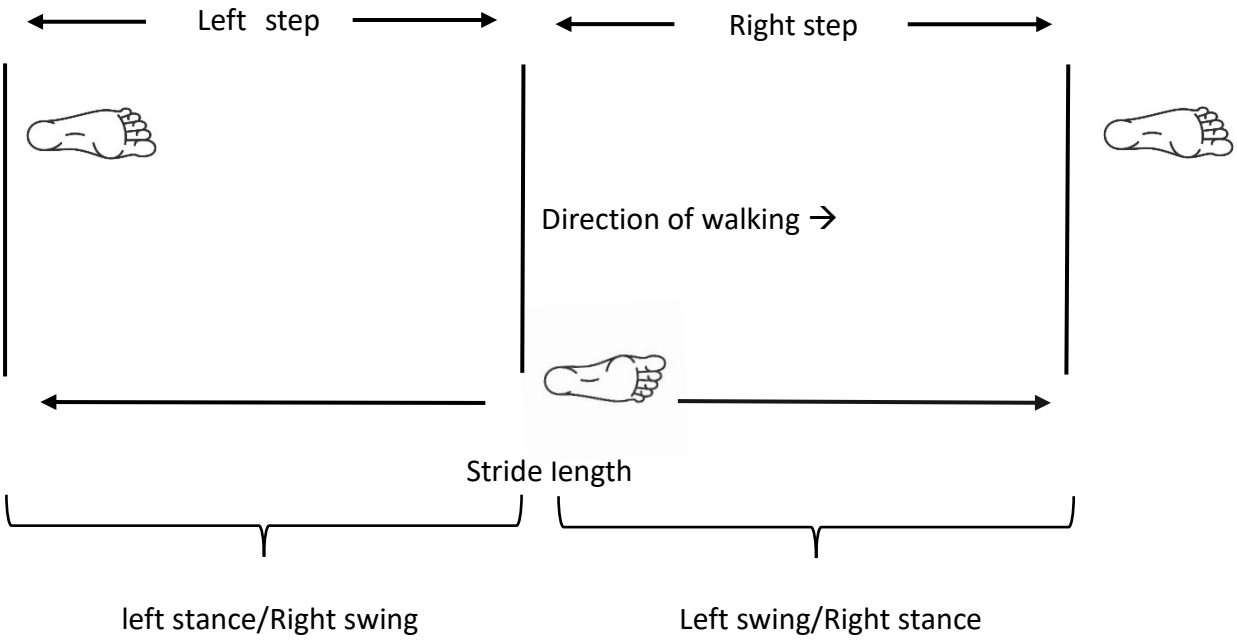


Figure 1. – Different parameters of a gait cycle. Modified from (Jessica Rose and Gamble 2006)

One gait cycle is divided into two main phases: Stance and swing (Figure 1). At a comfortable walking speed, and in populations without gait abnormalities, the percentage of the cycle during which the foot is on the ground in stance is approximately 62%, whereas during the swing, the time when the foot is in the air (i.e., not touching the floor) is 38% (Jessica Rose and Gamble 2006). The main subdivisions of gait phases, according to Rose and Gamble (2006), are Stance: 1) from foot strike to opposite foot-off (i.e., initial double limb support, 0-12%), 2) from opposite foot-off to opposite foot strike (i.e., single limb support, 12-50%), and 3) from opposite foot strike to foot-off (i.e., second double limb support, 50-62%), preparing the limb for swing. Subdivisions for Swing are 1) from foot-off to foot clearance (i.e., initial swing, 62-75%), 2) foot clearance to tibia vertical (i.e., mid-swing, 75-85%), and 3) from tibia vertical to foot strike (i.e., terminal swing, 85-100%) (Jessica Rose and Gamble 2006) (Figure 2).

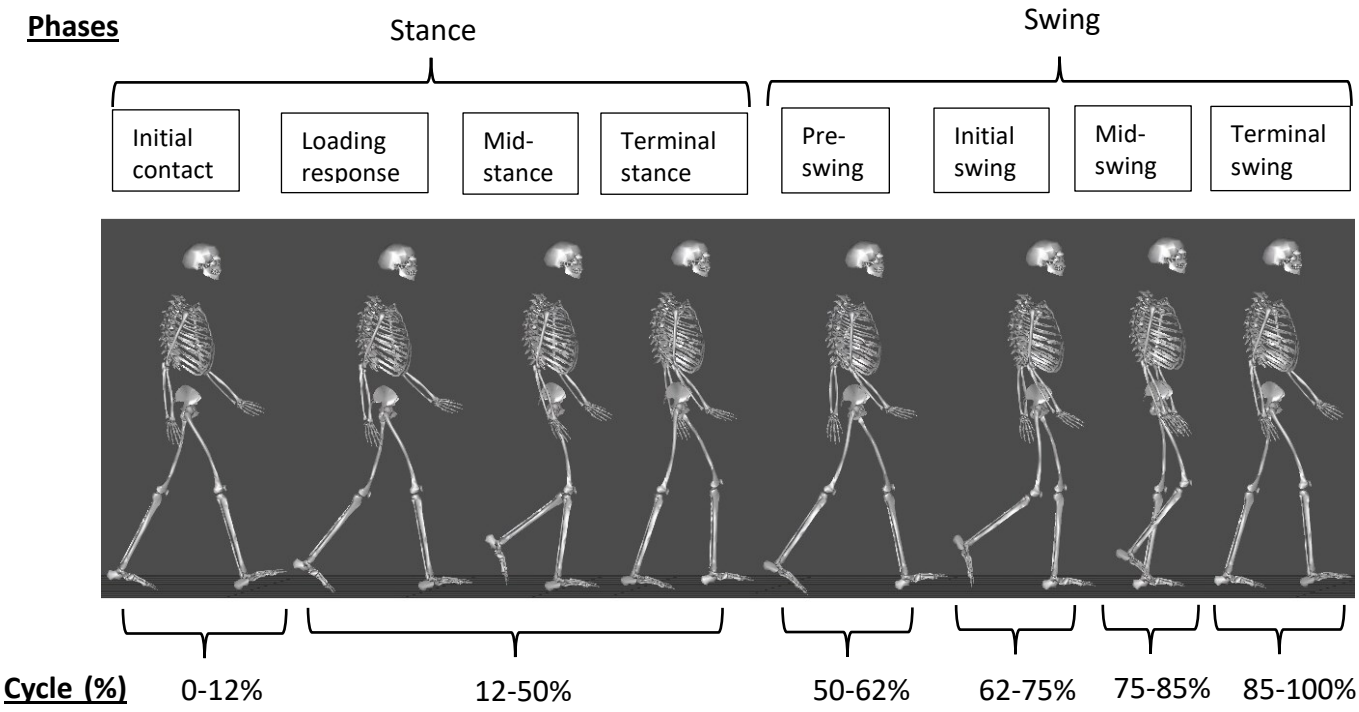


Figure 2. – Normal gait cycle. From first right foot strike to second right foot strike. It is modified from (P. Dixon 2015).

2.1.3 Muscular activation during gait

The muscular system plays a fundamental role in body movement. Muscles can adapt rapidly to any necessary change caused by either neural drive or external conditions (Gage et al. 2009). The combination of various muscle activities (e.g., agonist, antagonist) helps joints to move appropriately. Normal walking is produced by activating specific muscles, which contract mainly during stance or swing phases. Any out-of-phase or prolonged activation can indicate impaired motor control (e.g., CP, stroke) (Jessica Rose and Gamble 2006). Muscles mainly contract isometrically (i.e., isometric contraction) or while lengthening (i.e., eccentric contraction) during walking. These contractions help the body to save energy and maintain an upright posture when walking progresses (Jessica Rose and Gamble 2006).

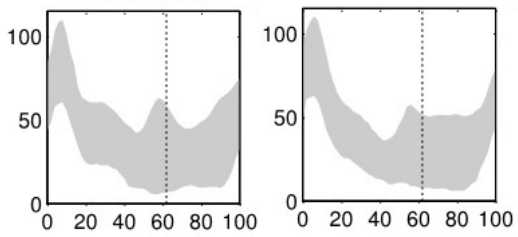
The main muscle activations during normal walking according to each gait phase are presented in the following paragraphs (Figure 3). The activation patterns are presented for: Gluteus Medius and Gluteus Maximums, which are hip abductor and extensor, respectively. Rectus Femoris (RF), Vastus Medialis (VM), and Vastus Lateralis (VL) help with knee extension, and Hamstrings (medial

and lateral) help with knee flexion. Also, Tibialis Anterior (TA) and Gastrecnemious (G) are ankle dorsiflexor and plantar flexor, respectively.

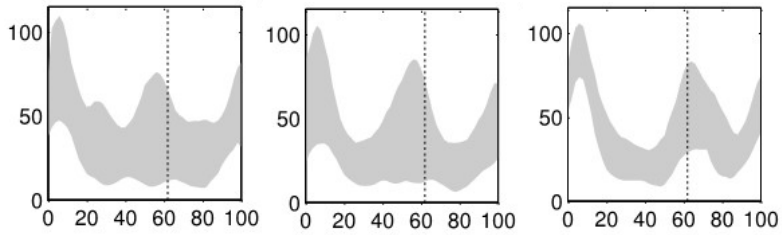
During stance, at initial contact, there is a simultaneous activation of the knee extensor and flexors to provide stability to the knee joint. In addition, ankle dorsiflexors contract eccentrically to prevent rapid descent of the foot on the floor (foot slap) (Jessica Rose and Gamble 2006). When the limb is in loading response, it accepts the body weight by eccentric contraction of the knee extensors, and then it starts to have concentric contraction to produce acceleration. At this moment, eccentric contraction of ankle plantar flexors helps knee extensors, leading to the movement of ground reaction force to the anterior part of the knee joint (i.e., to the forefoot) (Jessica Rose and Gamble 2006). In the meantime, isometric contraction of hip abductors contributes to pelvis stabilization. During mid-stance, eccentric contraction of the ankle plantar flexors helps the center of gravity to be at the highest point, and also, the knee remains extended without the need for the knee extensors contraction (Jessica Rose and Gamble 2006). At terminal stance, concentric contraction of ankle plantar flexors while the knee is passively extended leads to the body's power generation and forward movement (Jessica Rose and Gamble 2006).

Then the limb enters the swing phase. Pre-swing is the time during which the opposite limb accepts weight. In this phase, the hip flexors (i.e., iliopsoas and RF) produce concentric contraction to lift and swing the limb forward (Jessica Rose and Gamble 2006). In the swing phase, the passive pendulum behaviour of the limb leads to the passive movement at the following sub-phases. During the initial swing, there is no activation at the hip flexors. The leg is freely swinging (i.e., pendulum) at the knee joint, which affects the stride length and knee joint excursion. Also, the ankle dorsiflexors help the foot clearance by concentric contraction. This action of dorsiflexors along with the passive pendulum of the leg is continued during the mid-swing (Jessica Rose and Gamble 2006). Finally, at the terminal swing, knee flexors perform eccentric or isometric contraction to decelerate the limb, which extends the knee and bends the hip. Moreover, the isometric contraction of the ankle dorsiflexors helps the foot maintain a neutral position over the floor just before the next initial contact (Jessica Rose and Gamble 2006).

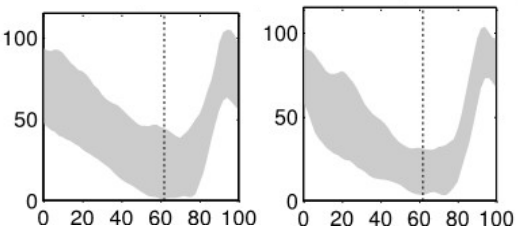
(a) Gluteus Medius (b) Gluteus Maximus



(c) Vastus Medial (d) Vastus Lateralis (e) Rectus Femoris



(f) Hamstrings Medial (g) Hamstrings Lateral



(h) Tibialis Anterior (i) Gastrocnemius

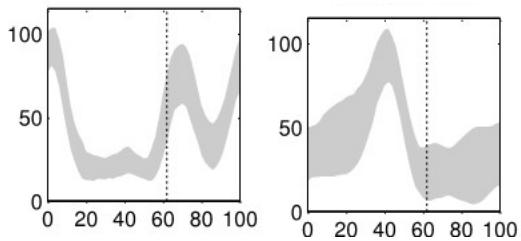


Figure 3. – Normal muscle activation of main muscles during gait. The Y axis is muscle activation level (0-100%), and X axis is a gait cycle (0-100%). The Grey area is one standard deviation from the mean, and vertical line is the beginning of the swing phase. With permission (P. Dixon 2015).

2.2 Muscle activity measurement

Electromyogram is an electrical signal associated with the contraction of a muscle (David A. Winter 2009a). Thanks to the study of EMG, fundamental information can be gained about the function of the muscles. For example, information regarding the muscle responsible for generating moments, muscle fatigue, and whether any antagonistic activation occurs (David A. Winter 2009a) can be quantified. Several factors, such as the shortening and lengthening velocity, tension, and fatigue, can affect the EMG signal (David A. Winter 2009a). Understanding the EMG's physiological and technological aspects help to interpret biomechanical adaptations appropriately (David A. Winter 2009a).

“Motor Unit” is the smallest unit describing the neural control of muscle contraction, which includes the cell body, dendrites, and axon, along with the corresponding muscle fibre (Konrad 2005) (Figure 4). Electrical potentials in the muscle fibres resulting from a motor unit's recruitment are known as motor unit action potential (David A. Winter 2009a). EMG electrodes on the surface (i.e., surface electrodes) or inside the muscles (i.e., indwelling electrodes) record the sum of all the motor unit action potentials carried to the muscle fibres at a time (David A. Winter 2009a). The choice of the electrode depends on the requirements of the investigation and research. For most kinesiological studies, surface electrodes are preferred as they are less invasive (Konrad 2005).

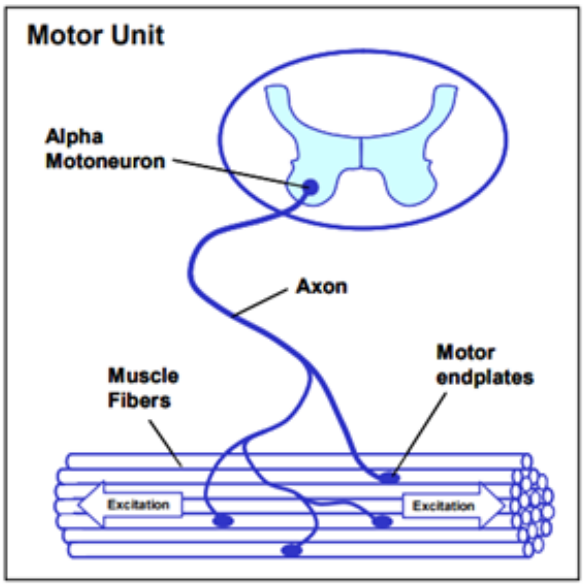


Figure 4. – Motor unit. With permission (Konrad 2005).

2.2.1 EMG acquisition

During the data acquisition process, some external sources of noise or artifacts impact the quality of the EMG signal. This mainly happens due to the sensitivity of the recording range, which starts from a few microvolts (Konrad 2005). For example, signals can be contaminated by baseline offset or shifts, which can be prevented if the EMG acquisition recommendations are made properly (Konrad 2005). Mainly, skin preparation (i.e., low skin impedance) and electrode placement (i.e., stable electrode) play key roles during the data collection process (Konrad 2005). Furthermore, the condition (e.g., static, dynamic) in which the data is collected should also be considered for skin preparation. For instance, very in-depth skin preparation is needed for more functional test conditions such as walking and running (Konrad 2005) due to the risk of movement artifacts. There are standard guidelines for EMG studies; Surface Electromyography for the Non-Invasive Assessment Muscles (SENIAM) guidelines, which is used during the signal acquisition, particularly in CGA (Hermens et al. 2000), and also International Society of Electrophysiology and Kinesiology guidelines which are used for analog and digital analysis (Meyer 1999). In addition to SENIAM recommendations for sensor placement, a muscle map has been identified by Konrad (2005) for

both surface and fine wire electrode placements, which are collected from other sources, showing the most common muscles investigated in kinesiological studies (Konrad 2005).

2.2.2 EMG signal processing

Raw EMG signals contain essential information about muscular excitation. Although an overall first understanding of the neuromuscular control is possible via qualitative assessment of the signals, a quantitative analysis would increase the reliability and validity of the results. In this way, EMG-specific signal processing steps are done in research (Konrad 2005). Konrad (2005) recommends that prior to any data analysis, it is important to check the validity and quality of the raw signals. Thus, before the signal processing, the following investigations could be done: 1) proof of the EMG-signal validity, 2) impedance Test, 3) inspection of the raw EMG-baseline quality, and 4) frequency distribution analysis.

After signal checking, EMG analysis can be done. The most common steps, according to the recommendations by Konrad (2005), are presented in this paragraph. In the first step, a bandpass filter is usually applied to the raw signal to remove low and high frequencies from the signal. According to the SENIAM recommendations for surface EMG, the cutoff frequency for the high pass is 10-20Hz, and it is “near 500 Hz” for the lowpass. The next step is “full wave rectification,” in which all the negative amplitudes are converted to positive amplitude so that the absolute value of the signal can be taken. Then “smoothing” is done by cutting the steep amplitude spikes in order to create the “linear envelope” of the signal (i.e., the mean trend of the signal). Common algorithms for this step are moving average, Root Mean Square (RMS), and Butterworth lowpass filtering. The fourth step is amplitude normalization to express the muscle activity between different muscles and individuals and across time in relation to a reference value obtained during standard conditions (A. Burden and Bartlett 1999). The reference value can be the internal mean, the peak value of a given trial, or the maximum voluntary activation obtained during a maximum voluntary contraction (MVC). The first presentation of EMG normalization was conducted by Eberhart et al. (1954) by normalizing the processed EMG signals from the quadriceps recorded during gait to a percentage of the maximum muscle activity that occurred during the gait cycle (Halaki et al. 2012). After amplitude normalization, “time normalization” is performed to average

the repetition of the movement. For example, in gait analysis, this method makes an equal amount of period (i.e., by collecting all the repetitions in a sequence), then calculates each period's mean value (Konrad 2005).

2.2.3 Muscle co-activation & co-contraction

As previously defined, MCa is the simultaneous activity of agonist and antagonist muscles crossing the same joint, which can increase joint stiffness (Diane L. Damiano 1993). It has been argued that MCa is a normal motor control strategy that individuals use based on the necessary task (e.g., complex activity, new task, an old task in a new condition) (T. S. Buchanan et al. 1986; Marsden, Obeso, and Rothwell 1983). The severity of MCa is influenced by the task requirements and the health situation of the individual (Carson and Riek 2001; Croce and Miller 2003; Enoka 1997). At the same time, Winter et al. (1978) have described MCo as the simultaneous contraction of the muscles around a joint "fighting" against each other without producing the "net movement," leading to inefficient movement (David A. Winter 2009a). MCo is considered one of the four causes of gait inefficiency (D. A. Winter and Robertson 1978), and it plays a part in developing motor skills in children (D. A. Winter 1979). To date, literature has used both terms (i.e., MCa, MCo) to study the action of the agonist and antagonist muscles around a joint during the walking trials.

Regarding the measurement, Falconer and Winter (1985) measured MCo using the moments of the force of agonist and antagonist muscles, $M_{agonist}$ and $M_{antagonist}$, respectively (Equation 2.1) (Falconer and Winter 1985). In addition, Winter (2005) suggested that if the profile of agonist and antagonist muscles activity during a specific movement is considered to measure the MCo, the common area of the activity of both muscles indicates the level of MCo (Equation 2.2) (David A. Winter 2005)

$$\%\, MCo = 2 \times \frac{M(antagonist)}{M(agonist)+M(antagonist)} \times 100 \quad \text{(Equation 2.1)}$$

$$\%MCo = 2 \times \frac{Common\ area\ of\ muscle\ 1\ \&\ muscle\ 2}{Area\ of\ muscle\ 1 + Area\ of\ muscle\ 2} \times 100 \quad \text{(Equation 2.2)}$$

There are numerous calculation methods for MCa in the literature, making it challenging to interpret the resulting MCa values and, in turn, understand the impact of MCa on movement

efficiency. These calculation methods include visual estimates of EMG magnitude, the ratio of EMG activities of agonist and antagonist muscles, and using normalized EMG to represent the antagonist muscles' activation levels (Ikeda et al. 1998). Busse et al. (2005) have summarized the existing MCa calculation methods, particularly concerning neurological populations, into four categories. Among those methods, only one of them was based on the quantification of the muscle moment. They take the view that the optimal method considers the antagonist activity and the contribution of the muscle to the joint moment (Busse, Wiles, and van Deursen 2005). Later a systematic review was conducted to recommend the best methodologies in MCo/MCa measurement (Rosa et al. 2014). This study included all the studies measuring the MCo/MCa in neurological populations, including CP during walking. According to this review, studies used different approaches to quantify the temporal MCo/MCa: 1) the time of overlap between the linear envelope of two opposite muscles, 2) the time of overlap between activity periods (onset delimited) of opposite muscles, the magnitude of MCo: 1) dividing the common area of the linear envelope of antagonist muscles by the sum of the areas of those muscles, 2) dividing the common area of the linear envelope between two muscles by the number of data points, and 3) calculating the difference between the minimum and maximum values of opposite muscles in each point of the gait cycle, and finally the amount of MCo: 1) the mean value of the area of overlap, 2) a correlation between the spectra of two opposite muscles, 3) quantification of the area of overlap between opposite muscles or 4) dividing this area by the overlap duration. Due to the considerable variability in MCo/MCa estimation methodologies, this study could not recommend the most appropriate methodology.

Knowing which factors affect the quality and the validity of MCo/MCa values would help evaluate the findings across studies. One factor is cross-talk between the muscles which is the amount of electrical activity produced by the neighbouring muscles (Konrad 2005). Although it has been shown that cross-talk between an agonist and antagonist pair is less than muscles of an agonist group (Kellis 1998), this factor should be considered while interpreting the MCo/MCa results. Another important factor is EMG normalization. In the previous section (Sub-section 2.2.2), we have described the common values that reference EMG normalization, all of which could result in various MCa levels and have various characteristics. For example, normalizing to MVC might be

problematic in studies in which the targeted muscle contraction type is different (e.g., concentric contraction) (Kellis 1998) or when the target population is children with neurologic impairments such as CP (Gagnat, Brændvik, and Roeleveld 2020). Thus, considering the method of EMG normalization might clarify the validity and reliability of the resulting MCa values. Finally, the importance of the role of muscle moments, especially in people with weak muscles, should be taken into consideration when calculating the MCo/MCa (Ikeda et al. 1998). If the MCo/MCa is quantified only with EMG activity without considering the EMG-moment relationship, the effects of the antagonist's muscle could be underestimated (Ikeda et al. 1998). Using only EMG to measure MCo/MCa is known to be conservative (Busse, Wiles, and van Deursen 2005). However, using this conservative method could be a constraint in assessing the dynamic activities, which might involve the activation of the bi-articular muscles and interaction between joints (i.e., regarding the moments) (Busse, Wiles, and van Deursen 2005). Overall, the major constraint among MCo/MCa calculation methods is the hypothesis that surface EMG can be used instead of muscle force. In particular, this could not be accepted during the concentric, dynamic movements as the EMG-force relationship is not linear (Kellis 1998).

2.3 Cerebral palsy

2.3.1 Background

CP is known as a group of disorders resulting from a non-progressive injury to a developing brain (McIntyre et al. 2011). Individuals with CP have a complex motor disorder related to the primary (e.g., muscle spasticity, loss of selectivity) and secondary deficiencies (e.g., muscle contractures, bony deformities). This population suffers from motor disorders that limit their movements and activities (e.g. walking and running) (Armand, Decoulon, and Bonnefoy-Mazure 2016). Particularly, in children with CP, ambulation is more impaired when the musculoskeletal system of the lower limb is more affected than the upper limb (Prosser et al. 2010), affecting their walking performance.

2.3.2 Types

Various classifications can distinguish the severity and type of impairment in the population with CP. CP can be classified according to the physiological characteristics which are related to the motor impairment (e.g., spastic, hypertonia, and hypotonia), the area of cerebral dysfunction (e.g., pyramidal or extrapyramidal), and the affected body segment (e.g., all four extremities, both legs, or one side of the body) (Gorter et al. 2004). Moreover, Miner (1956) has described a more comprehensive classification for the topographical aspect. The classification is: 1) principal involvement of one lower limb (i.e., monoplegia), 2) involvement of both lower limbs, and minimal involvement of upper limbs (i.e., diplegia), 3) involvement of one side of the body and the upper extremity is more involved (i.e., hemiplegia), 4) ipsilateral hemiplegia with contralateral monoplegia (i.e., triplegia), 5) involvement of all four limbs and the involvement of lower extremities is more (i.e., quadriplegia), 6) involvement of all four limbs and upper extremities are more involved (i.e., double hemiplegia) (Minear 1956). Another classification system is Gross Motor Function Classification System (GMFCS), in which Children with CP are classified into five levels according to their functional capacities at different ages (Rosenbaum et al. 2002). This system describes the functional characteristics of children with CP between the ages 6 and 12 years: level I) walking without restrictions and limitations in more advanced gross motor skills, level II) walking without assistive devices and limitations in walking outdoors and in the community, level III) walking with assistive mobility devices, and limitations in walking outdoors and in the community, level IV) limited self-mobility, and children are transported or use power mobility, level V) sever limited self-mobility even with using assistive technology (Rosenbaum et al. 2002).

2.3.3 Gait patterns

Changes in neural drive and muscles cause different motor functions in individuals with CP (Mathewson and Lieber 2015). For example, in spastic CP, the size of the muscle fibres varies significantly (Gage et al. 2009). It has been confirmed that muscle fibres in the affected limb are smaller than that of the non-affected limb in children with CP (Mohagheghi et al. 2007). Also, reduced strength resulting from the smaller muscle fibre in children with CP has been confirmed

by other researchers (Elder et al. 2003; Morse et al. 2004). This muscular pathology could contribute to atypical walking patterns in these children.

Children with CP have serious difficulties during gait. A spastic gait pattern is seen during the walking of children with CP (Aisen et al. 2011). Therefore, spasticity management is one of the main factors in gait rehabilitation (Green and Hurvitz 2007). Moreover, there are fundamental gait abnormalities in children with CP that should be investigated so that experts can conduct optimized treatment approaches (Armand, Decoulon, and Bonnefoy-Mazure 2016). Various classifications have been identified for gait in children with CP considering several factors such as the number of joints, segments, and planes (Dobson et al. 2007). In the following paragraphs, the primary classifications for gait abnormalities are described.

Winter et al. (1987) introduced the first gait pattern classification in individuals with unilateral spastic CP based on the kinematic data in the sagittal plane (Winters, Gage, and Hicks 1987). This classification represents increasing involvement from the first group toward the last one. Later, it was updated by other impairments (e.g., deviations associated with the transverse plane and knee hyperextension) (Rodda and Graham 2001). Four groups of gait deviations are as follow: 1) a drop foot during the swing preventing the first rocker at initial contact. Weakness or insufficient activity of TA, as well as excessive activation of G and Soleus (So) muscles, are responsible for this abnormality (i.e., Drop foot), 2-A) drop foot during the swing and continuous plantarflexion, restricting the ankle dorsiflexion during the stance (i.e., True equinus), 2-B) hyper-extended knee along with the characteristics of the previous group. Associated muscular adaptation for the second group is the contracture of ankle plantar flexors (i.e., G and So) (i.e., True equinus/Recurvatum knee), group 3) deviations related to the second group, minimum knee flexion during the swing, hyper-flexed hip, and excessive lordosis (i.e., True equinus/Jump knee), 4) all the previous deviations plus limited hip (i.e., hip extension in terminal stance and increased anterior pelvic tilt in the stance) and knee motion (Rodda and Graham 2001).

Gait deviations in individuals with diplegia are more complex due to bilateral involvement. An asymmetrical involvement can be seen between two limbs in most patients with diplegic CP (Gage et al. 2009). Rodda and Graham (2001) classified gait abnormalities in bilateral spastic CP using

Sutherland's classification (i.e., gait abnormalities of the knee) (Sutherland and Davids 1993). According to this classification, abnormal groups have 1) plantar-flexed ankle along with extended knee and hip (i.e., True equinus), 2) flexed knee and hip, increased lumbar lordosis (i.e., Jump knee), 3) excessive knee and hip flexion during stance (i.e., Apparent equinus), and 4) excessive ankle dorsiflexion, hip, and knee flexion (i.e., Crouch gait) (Rodda and Graham 2001). Later, Rodda et al. (2005) added a group to include the deviations related to the asymmetric gait; Thus, group 5) has a combination of issues such as apparent equinus and jump knee. Clinicians could benefit from this classification system to determine the potential causes of gait pathologies and the proper treatment strategy.

2.3.4 Co-activation and co-contraction patterns

As explained in the previous section, muscle activation patterns and muscle structure are altered in people with CP. Consequently, these changes influence the pattern of simultaneous contraction of the agonist and antagonist muscles crossing a joint. It has been suggested that smaller muscle size, changes in agonist muscle activation, and co-activation patterns are the adaptations leading to weakness in individuals with CP (Hussain et al. 2014). Furthermore, during gait, children with CP experience, a coactivation of antagonistic leg muscles, accompanied by an enhanced stretch reflex of short latency and a reduction of EMG activity in the extensor muscles of the leg. This is mainly due to the unestablished maturation of the gait pattern and lack of normal development (Wiltrud Berger 1998a). Undoubtedly, pathological gait is a major cause of inefficient muscle activity patterns and abnormal MCo/MCa levels in the population with CP. As it is also stated by Winter (2009) that co-contraction is more pronounced in hemiplegia and spastic cerebral palsy (David A. Winter 2009a), it is crucial to further investigate this phenomenon particularly in children with CP and compare it with healthy peers.

2.4 Problem and specific objective

Besides the main ambiguity regarding using the terms MCo and MCa for the analysis of the simultaneous activation of agonist and antagonist muscles, studies have been conducted with various objectives and methodologies. Considerable variability in the methods used for MCo/MCa assessment during gait is seen across studies. For example, the literature contains studies

undertaken on different muscle groups (e.g., shank, thigh), walking settings (e.g., ground, treadmill), walking speeds, and duration. Understanding the contribution of each of these experimental settings to the resulting MCo/MCa values in people with neurologic impairments such as CP is critical. It has been shown that walking on the ground for a longer duration will increase MCo values (Knarr, Zeni, and Higginson 2012; Parvataneni et al. 2009), meaning that resulting MCo/MCa can be affected in different ways. These variabilities and missing knowledge limit a direct comparison between studies in which healthy groups are compared with individuals with CP. Identifying whether the difference between groups is due to the experimental settings and methodological approaches or if it is because of their impairments seems to be critical. In addition, it is unclear if the level of functionality (i.e., GMFCS), type of CP, and gait abnormalities should be considered while comparing individuals with CP and healthy groups. Finally, we are not aware if studies calculated MCo or MCa. As a result, quantifying the difference in MCo/MCa values between children with CP and their TD peers during gait has remained unclear.

Therefore, the objective of this master's project is to distinguish MCo/MCa between children with CP and their TD peers during gait. Two studies are conducted in order to achieve the objective: (1) to describe MCo/MCa during walking in individuals with CP via a scoping review of all the existing literature by which we answer our questions and find the research gaps, and (2) to compare, using insights gained in the first study, MCo/MCa during gait in children with CP and their TD peers. The results of the first and second studies are reported in chapter 3 and 4, respectively. The hypotheses are (1) included studies in the scoping review will describe MCo/MCa during gait in individuals with CP (non-directional) and (2) MCo/MCa will be increased in children with CP compared to their TD peers.

Chapter 3 – [Muscle co-contraction and co-activation in cerebral palsy during gait: A scoping review]

3.1 Author's contribution

This chapter presents the result of the first study. The manuscripts is written by Sahar Mohammadyari Gharehbolagh, Cloe Dussault-Picard, Denis Arvisais, and Philippe Dixon. The article was submitted to the journal of Gait & Posture in October 2022, and it is under the review process at this time. This study meets the requirements of the master project which is to describe muscle co-contraction (MCo)/muscle co-activation (MCa) during gait in individuals with Cerebral palsy (CP) across all existing literature. All the co-authors contributed to this study. Sahar Mohammadyari Gharehbolagh was involved in all the steps of conducting the scoping review, namely, the development of the research question, the screening steps, data analysis, the interpretation of the results, and the drafting of the manuscript. Cloe Dussault-Picard assisted in data screening and revision of the manuscript. Denis Arvisais contributed to the database search step at the beginning of the project, and Philippe Dixon, the lead researcher, was involved in all aspects of the project. All the authors approved the final version of the manuscript.

3.2 Abstract

Background: CP results from an injury to a developing brain. Muscle activation patterns during walking are disrupted in individuals with CP. Indeed, excessive MCo or MCa is one of the characteristics of pathological gait. Although some researchers have studied MCo/MCa in individuals with CP during gait, inconsistent results limit our understanding of the literature. Increased knowledge of MCo/MCa patterns in individuals with CP may help the development of improved gait management approaches.

Research question: This review aimed to summarize MCo/MCa patterns while walking in individuals with CP across the existing literature and compare them with their healthy peers.

Methods: This study followed the Joanna Briggs Institute guidelines and the PRISMA Extension for Scoping Reviews recommendations. The recommendations of the Preferred Reporting Items for Systematic Reviews and Meta-Analyses Extension for scoping Reviews statement were respected. The following databases were searched: MEDLINE (Ovid), EMBASE (Ovid), CINAHL Plus with Full Text (Ebsco), SPORTDiscus with Full Text (Ebsco), and Web of Science.

Results: Among 2,545 identified studies, 21 studies remained after screening. In total, 337 participants with CP and 249 healthy participants were included. MCo and MCa terminologies are used for describing simultaneous muscle activation; however, when measured by electromyography (EMG), MCa terminology should be preferred to facilitate interpretation. A wide range of MCo/MCa patterns has been found across studies using different methodologies (e.g., gait protocol and computation methods). Finally, most of the included studies confirm that MCa is increased in individuals with CP during walking compared to controls.

Significance: This review identified missing concepts and common limitations in the literature which could be addressed in future research, such as the association between MCo/MCa and gait deviations and the most appropriate MCo/MCa computation method.

Keywords: Cerebral palsy, Electromyography, Gait analysis, Muscle activity

3.3 Introduction

Cerebral Palsy (CP) is a neurodevelopmental condition resulting from a brain injury that causes movement and motor impairments (Aisen et al. 2011). In individuals with CP, balance and gait patterns are compromised due to muscle spasticity, altered motor control and muscle strength, and the resulting musculoskeletal deformities (Crenna 1998; Ostensjø, Carlberg, and Vøllestad 2004). Walking on varied surfaces at different speeds while controlling balance, energy generation, and absorption requires muscle group synchronization (David A. Winter 2009b). These complex muscle activation patterns can be quantified via electromyography (EMG) (Gage 1993).

Muscle co-contraction (MCo) or muscle co-activation (MCa) has been used interchangeably in the literature to describe the coincident contraction of agonist and antagonist muscles (Busse, Wiles,

and van Deursen 2005). Excessive or poorly controlled MCo/MCa is a significant cause of inefficient gait (D. A. Winter 1978) in several pathologies, especially in CP (David A. Winter 2009b). According to Falconer and Winter (1985), the precise role of MCo/MCa in skilled movements such as walking has not been clarified by the literature (Falconer and Winter 1985). However, few researchers have investigated it during gait in individuals with CP. Of these, studies were conducted via diverse experimental protocols (e.g., gait speed, environment) and diverse quantification techniques (e.g., the percentage of time of simultaneous activation or common area of linear envelop of an agonist/antagonist pair), which leads to an unclear description of MCo/MCa and its impact on gait performance in individuals with CP.

Thus, this scoping review aimed to describe MCo/MCa and its role in modulating gait performance in individuals with CP. This research will determine whether these individuals experience different levels of MCo/MCa compared to their healthy peers and which variables (e.g., gait speed, CP types) affect this phenomenon. This is the first study to summarize the current literature concerning MCo/MCa during gait in individuals with CP. Considering the contribution of excessive MCo/MCa to gait inefficiency (D. A. Winter 1978)~~,~~ this knowledge could help to improve the choice of interventions or treatments offered by clinicians.

3.4 Materials and Methods

This scoping review was conducted according to Joanna Briggs Institute framework (Peters et al. 2017). This framework was introduced by Arksey and O'Malley (2005) (Arksey and O'Malley 2005) and later amended by Levac et al. (2010) (Levac, Colquhoun, and O'Brien 2010). Also, this framework comprises the identification of a research question, identification of the relevant studies, study selection, charting of the data, summarizing, reporting the results, and consultation (optional). The last step was not applied in the current study. This scoping review also followed the recommendation reported by PRISMA Extension for Scoping Reviews (Tricco et al. 2018), excluding the optional items related to critical appraisal.

3.4.1 Protocol and registration

Before initiating the research, a protocol was registered with the Open Science Framework (registration DOI 10.17605/OSF.IO/9X6AJ).

3.4.2 Search strategy

First, the general research topic was searched across MEDLINE (Ovid) and CINAHL (Ebsco). We evaluated the current studies' titles, keywords, and related search terms in this primary search. Next, the third author (DA) conducted a secondary search using the specific terminologies of the keywords in the following databases: MEDLINE (Ovid), EMBASE (Ovid), CINAHL Plus with Full Text (Ebsco), SPORTDiscus with Full Text (Ebsco), and Web of Science Core Collection (Clarivate Analytics). The final search strategy for MEDLINE can be found in Appendix 1. After the last search conducted on April 9, 2021, the first author (SMG) checked all the references of the included studies to add more sources in case of availability. Duplicate citations were removed using EndNote (X9, Clarivate Analytics, PA, USA). The selected studies were uploaded to Covidence software (Melbourne, Victoria, Australia).

3.4.3 Study selection

The inclusion criteria for the studies were (1) written in English or French, (2) the whole population or part of the population included individuals with CP, (3) assessment of MCo or MCa during gait, (4) investigation of at least two lower limb agonist and antagonist muscle pairs. Articles without available full text were excluded. The year of the publication was not considered in the selection process. The first (SMG) and second (CDP) reviewers independently screened the titles and abstracts. In the second screening step, the same reviewers assessed the full text of all studies from the previous step. The two screeners discussed and solved possible conflicts; if required, the senior author (PCD) established a consensus.

3.4.4 Data charting process

During data extraction, tables were designed based on (1) general information (e.g., year of publication, authors name), participant's demographics (e.g., age, sex), protocol design (e.g., MCo/MCa calculation method) (Table 1), (2) MCo or MCa during walking in individuals with CP

compared to their healthy peers (Table 2), and (3) MCo or MCa during walking within individuals with CP and existing relationships with various gait parameters (Table 3).

3.5 Results

3.5.1 Results summary

Data interpretation was conducted using descriptive and numerical analysis. Authors were contacted for studies that did not report sample mean and standard deviation. As no responses were received, other summary metrics available (e.g., median or range) were reported. Then, all the information from the analysis of the included studies is presented to describe MCo/MCa in individuals with CP compared to their healthy peers during gait. Finally, we discuss the results and their implications for research and clinical practice.

3.5.2 Data retrieved

The search strategy identified 2,545 studies. The screening process is illustrated in the PRISMA flowchart (Page et al. 2021) (Figure.5). A total of 21 articles were included (W. Berger, Quintern, and Dietz 1982; D. L. Damiano et al. 2000; Dietz and Berger 1983; Gagnat, Brændvik, and Roeleveld 2020; Gross et al. 2013, 2015; Keefer et al. 2004; Kim, Bulea, and Damiano 2018, 2021; Leonard, Hirschfeld, and Forssberg 1991; Lorentzen et al. 2019; Maltais et al. 2004; Pinto et al. 2018; Prosser et al. 2010; Syczewska and Święcicka 2016; Tao et al. 2015; Unnithan, Dowling, Frost, and Bar-Or 1996; Unnithan, Dowling, Frost, Volpe Ayub, et al. 1996; M. Vinti et al. 2018; Wakeling, Delaney, and Dudkiewicz 2007; Zwaan, Becher, and Harlaar 2012).

Table 1 contains general information on the included studies. All studies were published between 1982-2021 and used observational study designs. Except for six studies (D. L. Damiano et al. 2000; Keefer et al. 2004; Maltais et al. 2004; Pinto et al. 2018; Syczewska and Święcicka 2016; M. Vinti et al. 2018), all studies included a control group (i.e., healthy peers). Concerning the population sample, 16 studies included only children (W. Berger, Quintern, and Dietz 1982; Csongradi, Bleck, and Ford 1979; D. L. Damiano et al. 2000; Gagnat, Brændvik, and Roeleveld 2020; Gross et al. 2013, 2015; Keefer et al. 2004; Kim, Bulea, and Damiano 2018, 2021; Leonard, Hirschfeld, and Forssberg 1991; Maltais et al. 2004; Pinto et al. 2018; Prosser et al. 2010; Syczewska and Święcicka

2016; Unnithan, Dowling, Frost, and Bar-Or 1996; Unnithan, Dowling, Frost, Volpe Ayub, et al. 1996; M. Vinti et al. 2018), four studies included both young adults (≤ 30.7 years old) and children (Dietz and Berger 1983; Lorentzen et al. 2019; Tao et al. 2015; Wakeling, Delaney, and Dudkiewicz 2007; Zwaan, Becher, and Harlaar 2012), and 1 study did not report participants' age (Dietz and Berger 1983). In total, 616 participants were included in this review (healthy; n = 249, CP; n = 337). The total number of individuals with CP might be less than 337 due to the potential overlap of participants across four studies (W. Berger, Quintern, and Dietz 1982; Dietz and Berger 1983; Unnithan, Dowling, Frost, and Bar-Or 1996; Unnithan, Dowling, Frost, Volpe Ayub, et al. 1996). All studies mention the CP type (hemiplegia: n= 162, diplegia: n= 125, quadriplegia: n= 4) except for 1 study (Zwaan, Becher, and Harlaar 2012). The Gross Motor Function Classification System (GMFCS) level was not specified in 8 studies (W. Berger, Quintern, and Dietz 1982; Dietz and Berger 1983; Gross et al. 2013; Leonard, Hirschfeld, and Forssberg 1991; Syczewska and Święcicka 2016; Unnithan, Dowling, Frost, and Bar-Or 1996; Unnithan, Dowling, Frost, Volpe Ayub, et al. 1996; Wakeling, Delaney, and Dudkiewicz 2007) and two studies reported only that individuals with different GMFCS levels (i.e., I, II, III) participated (M. Vinti et al. 2018; Zwaan, Becher, and Harlaar 2012); however, ten studies reported the number of participants for each level (GMFCS I; n = 92, GMFCS II; n = 52) (D. L. Damiano et al. 2000; Gagnat, Brændvik, and Roeleveld 2020; Gross et al. 2015; Keefer et al. 2004; Kim, Bulea, and Damiano 2018, 2021; Lorentzen et al. 2019; Maltais et al. 2004; Pinto et al. 2018; Tao et al. 2015). We converted the Gross Motor Function Measure (GMFM) to GMFCS (Marois et al. 2016) for 1 study to simplify our descriptive analysis (Maltais et al. 2004). The mean sample size for the CP group is 16.0 ± 10.9, with an average age of 9.5 ± 3.1 years (excluding one study reporting only the median age of 4 years old) (Syczewska and Święcicka 2016) and one study that did not report the age of participants (Dietz and Berger 1983), and the average sample size of 17.1 ± 10.9, with an average age of 11.6 ± 7.3 years for the healthy group (excluding one study reporting only the age range for the healthy group) (Zwaan, Becher, and Harlaar 2012). Gait trials were conducted on treadmills for nine studies (W. Berger, Quintern, and Dietz 1982; Dietz and Berger 1983; Keefer et al. 2004; Kim, Bulea, and Damiano 2021; Leonard, Hirschfeld, and Forssberg 1991; Lorentzen et al. 2019; Maltais et al. 2004; Unnithan, Dowling, Frost, and Bar-Or 1996; Unnithan, Dowling, Frost, Volpe Ayub, et al. 1996). All other studies used

a walkway (i.e., laboratory floor). Gait speed included self-selected, slow, and fast speeds, with eight studies quantifying the speed of walking (Table 1) (W. Berger, Quintern, and Dietz 1982; Dietz and Berger 1983; Keefer et al. 2004; Leonard, Hirschfeld, and Forssberg 1991; Lorentzen et al. 2019; Maltais et al. 2004; Unnithan, Dowling, Frost, and Bar-Or 1996; Unnithan, Dowling, Frost, Volpe Ayub, et al. 1996). Among all studies, five reported that their participants were assisted during walking (i.e., by an adult or assistive device) (Leonard, Hirschfeld, and Forssberg 1991; Lorentzen et al. 2019; Prosser et al. 2010; Tao et al. 2015; Wakeling, Delaney, and Dudkiewicz 2007) and three conducted both assisted and/or unassisted trials (Unnithan, Dowling, Frost, and Bar-Or 1996; Unnithan, Dowling, Frost, Volpe Ayub, et al. 1996; Zwaan, Becher, and Harlaar 2012). With respect to lower-limb muscle assessment, three studies measured only shank muscles (i.e., Tibialis Anterior (TA)/Gastrocnemius Medialis (GM), Soleus (So), Peroneus longus) (Dietz and Berger 1983; Lorentzen et al. 2019; M. Vinti et al. 2018), two only thigh muscles (i.e., Rectus Femoris (RF), Bicep Femoris (BF), Vastus Lateralis (VL), Medial Hamstring (MH)) (D. L. Damiano et al. 2000; Keefer et al. 2004), and the rest investigated both shank and thigh muscles.

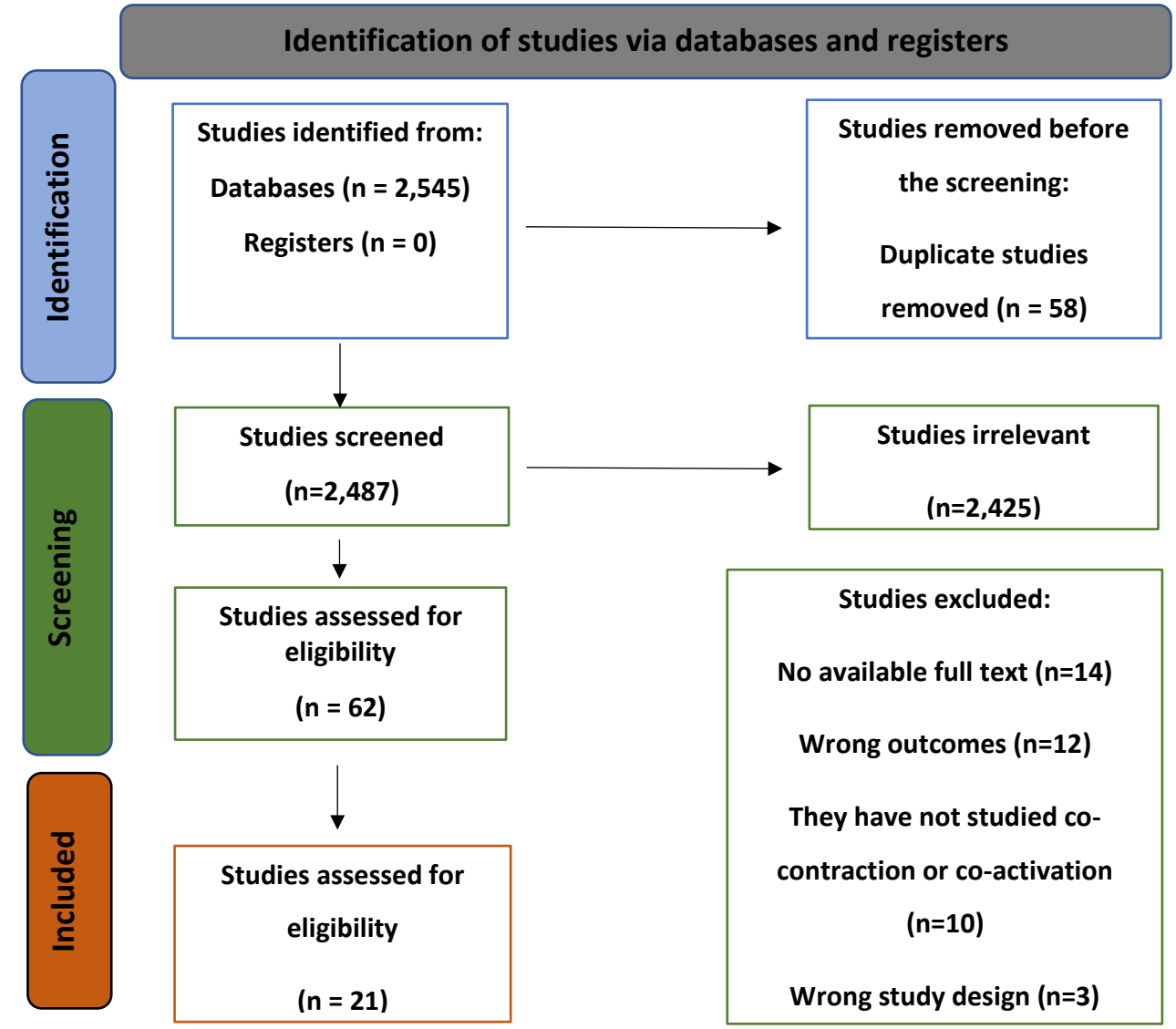


Figure 5. – PRIZMA flowchart for the selection process of studies

3.5.3 Calculation methods

Concerning terminology, nine studies used MCo (D. L. Damiano et al. 2000; Keefer et al. 2004; Leonard, Hirschfeld, and Forssberg 1991; Lorentzen et al. 2019; Pinto et al. 2018; Syczewska and Święcicka 2016; Unnithan, Dowling, Frost, and Bar-Or 1996; Unnithan, Dowling, Frost, Volpe Ayub, et al. 1996; M. Vinti et al. 2018), and 12 studies used MCa (W. Berger, Quintern, and Dietz 1982; Dietz and Berger 1983; Gagnat, Brændvik, and Roeleveld 2020; Gross et al. 2013, 2015; Kim, Bulea, and Damiano 2018, 2021; Maltais et al. 2004; Prosser et al. 2010; Tao et al. 2015; Wakeling, Delaney, and Dudkiewicz 2007; Zwaan, Becher, and Harlaar 2012), both to express concomitant muscle activity of agonist and antagonists muscles recorded by EMG.

Different computations were implemented to assess MCo/MCa across studies. Four studies expressed MCo or MCa as a percentage of the time of simultaneous activation of an agonist/antagonist pair based on EMG activity (W. Berger, Quintern, and Dietz 1982; Dietz and Berger 1983; Leonard, Hirschfeld, and Forssberg 1991; Prosser et al. 2010). Four studies (Gross et al. 2013, 2015; Pinto et al. 2018; Unnithan, Dowling, Frost, and Bar-Or 1996) conducted the method described by Unnithan et al. (1996) (Unnithan, Dowling, Frost, Volpe Ayub, et al. 1996), which represents the common area of the linear envelope in the agonist and antagonist pair divided by the number of data points. Two studies calculated cross-correlation coefficients across muscle pairs (Syczewska and Święcicka 2016; Zwaan, Becher, and Harlaar 2012). Two studies estimated the mean value of the area of overlap of the linear envelopes of the agonist/antagonist pair (D. L. Damiano et al. 2000; Maltais et al. 2004). Keefer et al. (2004) (Keefer et al. 2004) used the equation introduced by Falconer & Winter (1985) (Falconer and Winter 1985), which is the ratio between the antagonist muscle activity to the mean total muscle activity multiplied by two (see Table 1 for equation). Gagnat et al. (2020) used the same method as Keefer et al. (2004) (Keefer et al. 2004) and also quantified the antagonist activity as a percentage of agonist muscle activity (Gagnat, Brændvik, and Roeleveld 2020). Lorentzen et al. (2019) did not reference their method, which was the ratio between the root mean squared (RMS) EMG of one agonist and antagonist muscle (Lorentzen et al. 2019). Tao et al. (2015) studied the dynamic complexity of MCo using multivariate multi-scale entropy (Tao et al. 2015). One study used a method suggested by Vinti et al. (2013) (Maria Vinti et al. 2013), which represents the ratio of a muscle when acting

as an antagonist to the same muscle when acting as an agonist during a specific effort (M. Vinti et al. 2018). Finally, Wakelinga et al. (2007) evaluated MCa through wavelet decomposition of the EMG signals and principal component analysis (Wakeling, Delaney, and Dudkiewicz 2007). Kim et al. (2021) used non-negative matrix factorization to extract the synergy of the agonist and antagonist muscle pair (Kim, Bulea, and Damiano 2018, 2021).

3.5.4 MCo/MCa in individuals with CP vs. healthy peers

Specific agonist/antagonist muscular activity is summarized across studies in Tables 2 and 3.

Berger et al. (1982) reported higher MCa in CP group during the stance phase for the TA/GM and RF/BF muscle pairs compared to the healthy group (W. Berger, Quintern, and Dietz 1982). Diets and Berger (1983) also reported higher MCa in CP group during the stance phase for the TA/GM muscle pair (Dietz and Berger 1983). In one study conducted by Gagnet et al. (2020), two MCa calculation methods with (i.e., normalized) and without (i.e., non-normalized) EMG normalization were compared (Gagnat, Brændvik, and Roeleveld 2020). They found higher MCa (non-normalized) in terminal stance for TA/GM and TA/So muscle pairs and during weight acceptance for the TA/So pair in the CP group compared to the healthy group. Moreover, they reported higher MCa (normalized) during weight acceptance, mid-swing, and terminal swing for TA/GM and weight acceptance for TA/So. For the RF/MH muscle pair, higher MCa (normalized) during pre-swing in children with CP was reported. According to Leonard et al. (1991), the duration of MCo for TA/Lateral Gastrocnemius (LG) and RF/BF muscle pairs was increased in both CP and healthy groups during supported walking compared to independent walking (Leonard, Hirschfeld, and Forssberg 1991). Also, MCa for the TA/LG muscle pair significantly increased during independent walking in CP group. Finally, they observed that regardless of gait condition, MCa of the thigh muscles was higher than for the shank in the CP group. In addition, Prosser et al. (2010) found increased MCa duration for the RF/Semitendinosus (ST) muscle pair in the CP, compared to the healthy group (Prosser et al. 2010).

Tao et al. (2015) investigated MCa complexity for thigh and shank muscles (Tao et al. 2015). In their study, a CP group showed increased complexity during stance and decreased complexity during the swing. Moreover, complexity variability was higher in the CP group for all muscles.

Two studies reported higher MCo in CP rather than healthy group for TA/So and VL/MH muscle pairs across two walking speeds (Unnithan, Dowling, Frost, and Bar-Or 1996; Unnithan, Dowling, Frost, Volpe Ayub, et al. 1996). Both also showed that MCo was higher for the shank than the thigh at both speeds for both groups.

Among the three studies that compared MCa via muscle synergy, Kim et al. (2021) identified CP-specific clusters that showed excessive MCa values in the more affected limb for both thigh (RF/BF, VL/MH, RF/MH) and shank (TA/GM, TA/So, TA/PL) muscle pairs (Kim, Bulea, and Damiano 2021). On the other hand, in the study by Zwaan et al. (2012), the synergy of VM/ST muscle pair decreased in the CP compared to the healthy group (Zwaan, Becher, and Harlaar 2012).

Finally, Wakelinga et al. (2007) revealed that in RF/Semimembranosus and TA/GM muscle pairs, EMG-normalcy scores were significantly higher for the CP, compared to the healthy group during stance, swing, and across a full stride (Wakeling, Delaney, and Dudkiewicz 2007).

3.5.5 MCo/MCa within individuals with CP

3.5.5.1 Hemiplegic vs. diplegic

Increased MCa in individuals with diplegic, compared to hemiplegic CP for TA/GM and RF/BF muscle pairs, was reported by Berger et al. (1982) (W. Berger, Quintern, and Dietz 1982). This increase was immediately before and during stance (see Table 3).

3.5.5.2 Affected vs. non-affected leg

Syczewska & Święcicka (2016) reported greater MCo (i.e., agonist-antagonist correlation coefficients) of RF/MH muscle pair in the more affected hemiplegic leg than in less affected hemiplegic and diplegic legs (Table 3) (Syczewska and Święcicka 2016). There was a significant difference in MCo between the three groups (less affected hemiparetic legs, more affected hemiparetic legs, and diplegic legs) only for the RF/BF muscle pair. Moreover, Vinti et al. (2018) reported an increased MCo for TA/GM (during the mid and terminal swing) and TA/PL (during the initial, mid, and terminal swing) muscle pairs for the affected, compared to the non-affected limb during the mid and terminal swing (M. Vinti et al. 2018). Maltais et al. (2004) evaluated MCa during a 3-minute walking trial (Maltais et al. 2004). They reported decreased average MCa for

the Quadriceps/Hamstring pair in the more affected limb from minute 1 to minute 3 and greater average MCa at each minute in the more affected limb than in the less affected limb. For the TA/Triceps Surae muscle pair, MCa was decreased in less affected limbs, minute by minute and similarly between sides. Furthermore, MCa around the anckle was lower than around the knee during each minute. Finally, Kim et al. (2018) evaluated the synergy activation specific to individuals with CP and reported a higher level of MCa of RF/MH in the less affected leg (Kim, Bulea, and Damiano 2018).

3.5.5.3 Gait condition

In a comparison between independent and supported walking (adult-assisted), increased MCo for RF/BF and lower MCo for TA/LG muscle pairs have been reported in independent walking in the study by Leonard et al. (1991) (Leonard, Hirschfeld, and Forssberg 1991). Regarding gait speed, Gross et al. (2013) reported greater MCa for TA/So, RF/ST, and VM/ST muscle pairs when gait speed was increased, and this increase was higher for the affected compared to the non-affected limb (Gross et al. 2013). Also, according to the study by Unnithan et al. (1996), MCo of TA/So and VL/MH muscle pairs was greater when gait speed was increased (Unnithan, Dowling, Frost, Volpe Ayub, et al. 1996). They conducted another study, reporting that MCo of the same muscle pairs was the major factor in increasing the energy cost of walking only in children with CP (Unnithan, Dowling, Frost, and Bar-Or 1996). Based on a regression model identified by Pessoa et al. (2018), MCo of the VL/BF muscle pair was one of the predictors of gait speed (Pinto et al. 2018). Another model identified MCo of the same muscle pair as the only predictor of metabolic cost during gait. Finally, Damiano et al. (2000) reported that MCo of RF/BF had an inverse relationship with energy index expenditure (EEI) and was higher during fast gait than free gait and higher during gait than during isometric contractions (D. L. Damiano et al. 2000).

3.5.5.4 Joint motion

Gross et al. (2015) reported a negative relationship between MCa of TA/So muscle pair and the excursion of ankle plantarflexion during push-off and ankle dorsiflexion during the swing (Gross et al. 2015). Increased MCa for the RF/ST and VM/ST muscle pairs was also related to less joint excursion during knee loading and greater excursion of hip extension for the RF/ST muscle pair.

Table 1 – General characteristics of studies

	Title	Authors	Year	Demographics					Protocol design			
				n	Age	Sex	CP type	GMFCS Level (n)	Trials (gait condition; speed; shod/barefoot; assistance in gait)	EMG acquisition (SENIAM guidelines; processing; normalization)	Quantification methods	Assessed muscle (Pairs or single)
1	Pathophysiology of gait in children with cerebral palsy	Berger, Quintern, Dietz	1982	CP: 10 HC:10	CP: 12 HC: 11	CP:8F/2MHC: N/A	H:4 D: 6	N/A	Treadmill; 1-2 km/h; N/A; unassisted	N/A; rectification, averaging, smoothing; N/A	MCa: N/A	GM/TA, RF/BF
2	Muscle force production and functional performance in spastic cerebral palsy: relationship of co-contraction	Damiano, Martellotta, Sullivan et al.	2000	CP: 10	CP: 9.2	N/A	H: 1 D: 9	Level I: 3 Level II: 2 Level III: 5	Walkway; self-selected and fast; barefoot; unassisted	No; low-pass RMS filter; to MVC	MCo: a. Minimal EMG / maximal EMG at each time point b. Mean value of the area of overlap of the linear envelopes of the agonist/antagonist pair	RF/BF
3	Normal and impaired regulation of muscle stiffness in gait: A new hypothesis about muscle hypertonia	Diets, Berger	1982	CP: 10 HC: 10 HA:20	N/A	N/A	D: 6	N/A	Treadmill; 1-2 km/h; shod; unassisted	N/A; Rectification, averaging; N/A	MCa: N/A	GM/TA
4	Surface electromyography normalization affects the interpretation of muscle activity and coactivation in children with cerebral palsy during walking	Gagnat, Brændvik, Roeleveld	2020	CP: 23 HC: 11	CP: 11.7 HC: 9.4	CP: F/16M HC:7F/4M	H: 17 D: 6	Level I: 17 Level II: 6	Walkway; self-selected; barefoot; unassisted	Yes; band-pass filter, RMS; to maximum value	MCa: a. 2 * (antagonist/ agonist+antagonist) *100 b. (antagonist/agonist) *100	TA/SO, TA/GM, RF/HM

	Title	Authors	Year	Demographics					Protocol design			
				n	Age	Sex	CP type	GMFCS Level (n)	Trials (gait condition; speed; shod/barefoot; assistance in gait)	EMG acquisition (SENIAM guidelines; processing; normalization)	Quantification methods	Assessed muscle (Pairs or single)
5	The influence of gait speed on co-activation in unilateral spastic cerebral palsy children	Gross, Leboeuf, Hardouin, et al.	2013	CP: 10 HC: 10	CP: 10.1 HC: 10.2	N/A	H:10	N/A	Walkway; self-selected/ fast/slow; barefoot; unassisted	Yes; rectification; filtering; to maximum value	MCa: Common area of linear envelop in agonist and antagonist pair divided by the number of data points	RF/ST, VM/ST, TA/SO
6	Does muscle coactivation influence joint excursions during gait in children with and without hemiplegic cerebral palsy? Relationship between muscle coactivation and joint kinematics	Gross, Leboeuf, Hardouin, et al.	2015	CP: 12 HC: 12	CP: 9.9 HC: 9.7	N/A	H: 12	Level I: 12	Walkway; self-selected/fast/slow; barefoot; unassisted	Yes; rectification, filtering; to maximum value	MCa: Common area of linear envelop in agonist and antagonist pair divided by the number of data points	RF/ST, VM/ST, TA/SO
7	Interrelationships among thigh muscle co-contraction, quadriceps muscle strength and the aerobic demand of walking in children with cerebral palsy	Keefer, Chen, Liou	2001	CP: 13	CP: 11.2	CP: 5F/8M	H: 13	Level I: 12 Level II: 1	Treadmill; 0.04, 0.05, 0.06 km/h; barefoot; unassisted	Yes (partial); rectification, low-pass filter; to mean value	MCo: 2 * (common area of agonist and antagonist/ (area of agonist + area of antagonist)) * 100	VL/MH
8	Greater Reliance on cerebral palsy-specific muscle synergies during gait relates to poorer temporal-spatial Performance Measures	Kim, Bulea, Damiano	2021	CP: 10 HC: 10	CP: 14.9 HC: 15.0	CP: 7F/3M HC: F/3M	H: 10	Level I: 6 Level II: 4	Treadmill; Self-selected; N/A; N/A	Yes; high-pass filter, rectification, low-pass filter; to maximum value	Synergy: non-negative matrix factorization	TA, EH, LG, SO, RF, VL, ST, BF

	Title	Authors	Year	Demographics					Protocol design			
				n	Age	Sex	CP type	GMFCS Level (n)	Trials (gait condition; speed; shod/barefoot; assistance in gait)	EMG acquisition (SENIAM guidelines; processing; normalization)	Quantification methods	Assessed muscle (Pairs or single)
9	Children with cerebral palsy have greater stride-to-stride variability of muscle synergies during gait than typically developing children: implications for motor control complexity	Kim, Bulea, Damiano	2018	CP: 20 HC: 8	CP: 12.5 HC: 12.0	CP: F/11M HC: F/3M	H: 17 D: 3	Level I: 12 Level II: 8	Walkway; self-selected; N/A; N/A	Yes; high-pass filter; to maximum value	Synergy: non-negative matrix factorization	TA, GM, RF, MH
10	The development of independent walking in children with cerebral palsy	Leonard, Hirschfefd, Forssberg	1991	CP: 8 HC: 5	CP:2.5 HC: 1.15	N/A	D: 2 H:6	N/A	Treadmill; self-selected, 1.44 km/h; barefoot; assisted	N/A; band-pass filter; N/A	MCo: time of overlap between activity periods of gonist/antagonist pair	TA/LG, RF/BF
11	Maturation of feedforward toe walking motor program is impaired in children with cerebral palsy	Lorentzen, Willerslev Olsen, Hüche Larsen, et al.	2019	CP: 28 HC: 24 HA: 15	CP: 7.0 HC: 6.5 HA: 30.7	N/A	H:16 D:12 Q: 1	Level I: 15 Level II: 8 Level III: 5	Treadmill; self-selected, 1.1 ± 0.3km/h; barefoot; assisted	No; band-pass filter; N/A	MCo: Ratio between agonist and antagonist pair	TA/SO
12	Minute by minute differences in co-activation during treadmill walking in cerebral palsy	Maltais Pierrynowski, Galea, et al.	2004	CP: 8	CP: 11.9	CP: 3F/5M	H: 3 D: 5	Level I: 5 Level II: 3	Treadmill; 3.6±1.9 km/h; shod; unassisted	Yes (partial); rectification, low-pass filter; to maximum value	MCa: Mean value of the area of overlap of the linear envelopes of the agonist/antagonist pair	Qu/H, TA/TS
13	Mechanisms contributing to gait speed and metabolic cost in children with unilateral cerebral palsy	Pessoa, Pinto, Teixeira Fonseca, et al.	2018	CP:14	CP: 7.8	CP: 5F/9M	H: 14	Level I: 6 Level II: 8	Walkway; self-selected; barefoot; unassisted	N/A; rectification, band-pass filter; to MVC	MCo: Common area of linear envelop in agonist/ antagonist pair divided by the number of data points	GMx/RF, VL/BF, TA/LG

	Title	Authors	Year	Demographics					Protocol design			
				n	Age	Sex	CP type	GMFCS Level (n)	Trials (gait condition; speed; shod/barefoot; assistance in gait)	EMG acquisition (SENIAM guidelines; processing; normalization)	Quantification methods	Assessed muscle (Pairs or single)
14	Trunk and hip muscle activation patterns are different during walking in young children with and without cerebral palsy	Prosser, Lee, VanSant, et al.	2010	CP: 15 HC: 16	CP: 5.2 HC: 3.3	CP: F/10M HC: F/7M	D: 14 Q: 1	Level II:7 Level III: 8	Walkway; self-selected barefoot; assisted	Yes; low-pass filter, averaging; N/A	MCa: time of simultaneous activation of agonist and antagonist pair	ES/RA, RF/ST
15	Are electromyographic patterns during gait related to abnormality level of the gait in patients with spastic cerebral palsy?	Syczewska, Święcicka	2016	CP: 51	CP: 4 (median)	N/A	H: 26 D: 25	N/A	Walkway; self-selected; N/A; unassisted	Yes; rectification, low-pass filter, averaging; N/A	MCo: Averaged cross-correlation coefficient of agonist and antagonist pair	RF/LH, RF/MH, TA/LG
16	Multi-scale complexity analysis of muscle coactivation during gait in children with cerebral palsy	Tao, Zhang, Chen, et al.	2015	CP: 11 HC: 8 HA: 7	CP: 5.4 HC: 6.6 HA: 24.7	CP:2F/9F HC:5F/3M HA:7M	H: 5 D: 6	Level I: 4 Level II: 5 Level III: 2	Walkway; self-selected; N/A; assisted	No; N/A; N/A	MCa (dynamic complexity of): multivariate multi-scale entropy (MMSE)	TA/ SO/ LG, VL/ RF/ SE/ BF/ TF
17	Role of co-contraction in the O2 cost of walking in children with cerebral palsy	Unnithan, Dowling, Frostet al.	1996	CP: 9 HC: 9	CP: 12.7 HC: 13.6	CP: 2F/7M HC: F/7M	H: 1 D: 7 Q: 1	N/A	Treadmill; 3 km/h and 90% FWS; N/A; assisted and unassisted	Yes (partial); rectification, low-pass filter; to maximum value	MCo: Common area of linear envelop in agonist and antagonist pair divided by the number of data points	VL/MH, TA/SO
18	Co-contraction and phasic activity during gait in children with cerebral palsy	Unnithan, Dowling, Frost, et al.	1996	CP: 9 HC: 8	CP: 12.7 HC: 13.6	CP: 2F/7M HC: F/7M	H: 1 D: 7 Q: 1	N/A	Treadmill; 3 km/h and 90% of FWS; N/A; assisted and unassisted	Yes (partial); rectification, low-pass filter; to maximum value	MCo: Common area of linear envelop in agonist/ antagonist pair divided by the number of data points	VL/MH, TA/SO

	Title	Authors	Year	Demographics					Protocol design			
				n	Age	Sex	CP type	GMFCS Level (n)	Trials (gait condition; speed; shod/barefoot; assistance in gait)	EMG acquisition (SENIAM guidelines; processing; normalization)	Quantification methods	Assessed muscle (Pairs or single)
19	Muscle shortening and spastic cocontraction in gastrocnemius medialis and peroneus longus in very young hemiparetic children	Vinti, Bayle, Merlo, et al.	2018	CP: 10	CP: 3	CP: 4F/6M	H: 10	Levels I or II	Walkway; self-selected; barefoot; unassisted	Yes; high-pass filter, low-pass filter, rectification; MVC	MCo: RMS of a muscle while acting as an antagonist/RMS of same muscle while acting as an agonist	GM-PL/TA
20	A method for quantifying dynamic muscle dysfunction in children and young adults with cerebral palsy	Wakelinga, Delaneyb, Dudkiewicz	2007	CP: 17 HC+HA: 36	CP: 11.3 HC +HA: 10.8	N/A	D: 17	N/A	Walkway; self-selected; N/A; assisted	No; N/A; N/A	MCa: Correlation spectra analyzed between two antagonist muscles	RF/SM, TA/GM
21	Synergy of EMG patterns in gait as an objective measure of muscle selectivity in children with spastic cerebral palsy	Zwaan, Becher, Harlaar	2012	CP: a. (39) b. (38) HC+HA: 30	CP: a. (6.5) b. (N/A) HC+HA: 3-20 (range)	N/A	N/A	Levels I-III	Walking; self-selected and slow; barefoot; assisted or unassisted	Yes; rectification, low-pass filter; N/A	Synergy: Cross correlations between the envelopes of the ensemble average EMG profiles of selected muscles for each subject	VM/ST, VM/GM

Abbreviations: CP, cerebral palsy; HC, Healthy children; HA, healthy adults; H, hemiplegia; D, diplegia; P, paraplegia; Q, quadriplegia; M, male; F, female; GMFCS, gross motor functional classification system; FWS, fastest walking speed; EMG, electromyography; SENIAM, Surface Electromyography for the Non-Invasive Assessment Muscles; MVIC, maximal voluntary isometric contraction; RMS, root mean square; MCa, muscle co-activation; MCo, muscle co-contraction; G, gastrocnemius; TA, tibialis anterior; RF, rectus femoris; BF, biceps femoris; VM, vastus medialis; MH, medial hamstrings; GM, gastrocnemius medialis; SO, soleus; ST, semi-tendinosis; VL, vastus lateralis; EH, extensor hallucis longus; LG, lateral gastrocnemius; GMx, gluteus maximus; Qu, quadriceps; H, hamstrings; TS, triceps surae; TZ, trapezius; ES, erector spinae; RA, rectus ab-dominis, EO, external oblique; GMd, gluteus medius; PL, peroneus longus; TFL, tensor fasciae lata; SM, semimembranosus.

Table 2 - MCo/MCa in individuals with CP, compared to their healthy peers

MCo/ MCa (Muscle pair)	Significant differences in CP
TA/GM	↑ immediately before and during the stance (1) ↑ stance (1, 3) ↑ terminal stance (abs) (4) ↑ weight acceptance, mid-swing, terminal swing (norm) (4) ↑ in more affected limb (synergy) (8) ↑ (variation of complexity) (16) ↑ stance, right leg (complexity) (16) ↓ swing, right leg (complexity) (16)
TA/SO	↑ terminal stance (abs) (4) ↑ weight acceptance (norm) (4) ↑ in more affected limb (synergy) (8) ↑ (variation of complexity) (16) ↑ stance, right leg (complexity) (16) ↓ swing, right leg (complexity) (16) ↑ (17,18)
TA/LG	↑ (duration) (10) ↑ (variation of complexity) (16) ↑ stance, right leg (complexity) (16) ↓ swing, right leg (complexity) (16)
TA/PL	↑ in more affected limb (synergy) (8)
RF/BF	↑ immediately before and during stance (1) ↑ in more affected limb (synergy) (8) ↑ (duration) (10) ↑ (variation of complexity) (16) ↑ stance, right leg (complexity) (16)
VL/MH	↑ in more affected limb (synergy) (8) ↑ (17,18)
RF/MH	↑ (abs, norm) (4) ↑ pre-swing (norm) (4) ↑ in more affected limb (synergy) (8)
RF/ST	↑ (duration) (14) ↑ (variation of complexity) (16) ↓ swing, right leg (complexity) (16)
VL/BF	↑ (variation of complexity) (16) ↑ stance, right leg (complexity) (16)
VL/ST	↑ (variation of complexity) (16) ↑ stance, right leg (complexity) (16)
VM/ST	↓ synergy (21)

Abbreviations (study numbers are based on Table 1): TA, tibialis anterior; GM, gastrocnemius medialis; SO, soleus; LG, lateral gastrocnemius; PL, peroneus longus; RF, rectus femoris; BF, biceps femoris; VL, vastus lateralis; MH, medial hamstrings; VM, vastus medialis; ST, semi-tendinosis; MCa, muscle co-activation, CP, cerebral palsy; H, healthy. ↑, increased/higher; ↓ decreased/lower.

Table 3 - Significant relationships of gait MCo/MCa with various outcomes in individuals with CP

MCa or MCo (Muscles pair)	Relationships
TA/GM	H < D (1) Affected limb > non-affected limb during mid and terminal swing (19)
TA/SO	↑ with gait speed; affected limb > non-affected limb (5) ↑ with gait speed (18) ↓ ankle plantarflexion during push-off (6) ↓ ankle dorsiflexion during swing in the affected limb (6) ↑ energy cost of walking (17) ↑ than thigh muscles (VL/MH) (17)
TA/LG	Supported walking > independent walking (10) ↓ than thigh muscles (RF/BF) (10)
TA/PL	affected > non-affected limb during mid and terminal swing (19)
TA/TS	↓ in the less affected limb (12) ↓ than thigh muscles (Qu/H) (12)
RF/BF	H < D (1) ↓ with EEI (2) Free gait < fast gait (2) Isometric test < gait (2) Supported walking < independent walking (10) More affected hemiplegic limbs > less affected hemiplegic and diplegic limbs (15)
VL/MH	↑ with speed (18) ↑ energy cost of walking (17)
RF/MH	↑ synergy: in the less affected limb (9) More affected hemiplegic limbs > less affected hemiplegic and diplegic limbs (15)
RF/ST	↑ with gait speed; affected limb > non-affected limb (5) ↓ joint excursion (knee loading) (in affected limb) (6) ↑ hip extension (in affected limb) (6)
VL/BF	A predictor of gait speed (13)
VM/ST	↑ with gait speed; affected limb > non-affected limb (5) ↓ joint excursion during knee loading (in the affected limb) (6)
Qu/H	↓ in the more affected limb (12) More affected limb > less affected limb (12)

Abbreviations (study numbers are based on Table 1): TA, tibialis anterior; GM, gastrocnemius medialis; SO, soleus; LG, lateral gastrocnemius; PL, peroneus longus; RF, rectus femoris; BF, biceps femoris; VL, vastus lateralis; MH, medial hamstrings; VM, vastus medialis; ST, semi-tendinosis; Q, quadriceps; H, hamstrings; H, hemiplegia; D, diplegia; MCa, muscle co-activation, CP, cerebral palsy; GMFCS, gross motor functional classification system; EEI, energy expenditure index; VO_2, maximal oxygen consumption; ↑, increased/higher; ↓ decreased/lower.

3.6 Discussion

3.6.1 Summary

This scoping review identified 21 studies investigating MCo/MCa during gait in individuals with CP and their healthy peers. Studies implemented a wide range of methods to assess gait, analyze EMG, and quantify MCo/MCa, restricting the comparability of results. Nonetheless, this scoping review enabled us to summarize trends of MCo/MCa and its role in gait for individuals with CP. Our analysis demonstrates that MCo/MCa is generally increased in the CP group compared to

controls. Also, this scoping review identifies CP-specific MCo/MCa patterns and highlights some of the gaps and inconsistencies with regard to the contribution of MCa on various factors (e.g., cost of walking, gait efficiency, and gait speed).

3.6.2 Co-contraction vs. co-activation

One of the major disparities in the assessed studies is the terminology used to express simultaneous agonist and antagonist muscle activation. Indeed, it has been stated that MCa and MCo can be used interchangeably when describing the concomitant contraction of agonist and antagonist muscles (Busse, Wiles, and van Deursen 2005); however, Ikeda et al. (1998) stressed that a distinction must be made regarding these two terminologies: MCa quantifies the co-activation of muscles based on EMG activity whereas MCo is defined as the percentage of the net moment that was negated by the antagonist moment (Ikeda et al. 1998). Among studies in this scoping review, 9 used MCo terminology (Table 1); however, none represent MCo as a percentage of the net moment but rather as a co-activation of muscles based on EMG activity. Thus, to avoid misuse of the MCo terminology, we have used the term MCa to describe the results in the discussion. Also, we suggest implementing a distinction between the two terminologies, as suggested by Ikeda et al. (1998).

3.6.3 Calculation methods

Our study confirms a wide range of calculation methods for MCa across studies. Most studies used two main methods: the common area of linear envelop in agonist and antagonist muscle pair and the time of simultaneous activation of an agonist/antagonist muscle pair. All other studies conducted different approaches. This finding is consistent with a previous systematic review, indicating that it is impossible to recommend the most appropriate methodology to evaluate MCa during gait in people with central nervous system disorders (Rosa et al. 2014). It is unclear whether this variability in methodologies is due to the different included CP types, gait protocols, EMG analysis, and processing or simply because of a lack of consensus on the MCa estimation. This is a crucial issue that should be addressed by future research.

3.6.4 MCa in individuals with CP vs. healthy peers

Studies that compared MCa between individuals with CP and a healthy group reveal that MCa around the ankle and/or around the knee is greater in individuals with CP during walking.

Berger et al. (1982) (W. Berger, Quintern, and Dietz 1982) and Dietz & Berger (1983) (Dietz and Berger 1983) suggested that increased MCa in individuals with CP, compared to their healthy peers, is a result of impaired maturation of the locomotor pattern. Their MCa results were consistent with MCa in stepping newborns (H 1980), indicating the contribution of disrupted maturation in the increased MCa for children with CP. It was also suggested that increased MCa might occur due to impaired supraspinal control or plasticity caused by the brain lesion (Wiltrud Berger 1998). The factors resulting from the brain injury, such as altered motor control and spasticity, may contribute to the increased MCa complexity that Tao et al. (2015) (Tao et al. 2015) reported, whereas muscle paralysis (Gormley 2001) and a lack of excitation-contraction coupling mechanisms may explain the decreased complexity during the swing. Indeed, it is known that the brain lesion causes an incomplete excitation of the superior motoneuron, leading to the activation of fewer motor units during voluntary contraction.

A phasic analysis is relevant to identifying gait abnormalities (Frigo and Crenna 2009) and understanding selective activation patterns during gait (Gagnat, Brændvik, and Roeleveld 2020); however, because studies partition their gait phases differently, the literature does not allow to summarize results via phasic analysis. For instance, Gagnat et al. (2020) divided the gait cycle into six phases (i.e., weight acceptance, mid-stance, terminal stance, pre-swing, initial swing, mid-swing/terminal swing) (Gagnat, Brændvik, and Roeleveld 2020), and Tao et al. (2015) divided the gait cycle into swing and stance phases (Tao et al. 2015). Thus, MCa results from the study by Gagnat et al. (2015) during weight acceptance (Gagnat, Brændvik, and Roeleveld 2020), which represents approximately 10% of the gait cycle (Perry, K, and Davids 1992) cannot be compared with results from Tao et al. (2015) during stance phase (Tao et al. 2015), which represents approximately 60% of the gait cycle.

According to three studies exploring MCa indices derived from synergy analysis (Kim, Bulea, and Damiano 2018, 2021; Zwaan, Becher, and Harlaar 2012), higher levels of MCa in individuals with

CP compared to their healthy peers is attributable to less selectivity (Kim, Bulea, and Damiano 2021), or muscle spasticity, tightness, or weakness (Kim, Bulea, and Damiano 2018). A third study by Zwaan et al. (2012) explored synergy but did not directly report co-activation data (Zwaan, Becher, and Harlaar 2012); however, the interpretation of their findings in terms of MCa showed that thigh synergy (i.e., VM/ST MCa) is lower in the CP compared to the healthy group. This is inconsistent with the results obtained from the two studies by Kim et al. (2018, 2021) (Kim, Bulea, and Damiano 2018, 2021), potentially due to the different synergy extraction methods. Zwaan et al. (2012) defined the synergy of muscle contraction based on cross-correlations between the envelopes of the ensemble average EMG profiles of selected muscles (Zwaan, Becher, and Harlaar 2012). That is, ‘‘thigh synergy’’ expresses the VM profile against the ST profile; however, in two studies conducted by Kim et al. (2018, 2021), non-negative matrix factorization was used (Lee and Seung 1999) to extract muscle synergies from EMG (included muscles shown in table 1) (Kim, Bulea, and Damiano 2018, 2021).

3.6.5 MCa within the CP group

3.6.5.1 Hemiplegic vs. diplegic

Among the two studies (W. Berger, Quintern, and Dietz 1982; Syczewska and Święcicka 2016) that compared MCa between CP types, both reported opposite results; Berger et al. (1982) reported higher MCa in diplegic legs, compared to the paretic leg of hemiplegic CP (W. Berger, Quintern, and Dietz 1982); whereas Syczewska and Święcicka (2016) reported an increased MCa for the more affected leg of hemiplegic participants compared to both legs of children with diplegic CP (Syczewska and Święcicka 2016). Both studies did not specify the GMFCS level of included individuals, and they conducted different methods to estimate MCa. Indeed, these differences may contribute to explaining these inconsistencies. It has been suggested that there is a clear correlation between the level of motor impairment (i.e., GMFCS level) and restricted motor development (Gage et al. 2009), which might affect the MCa levels in this population. Furthermore, different MCa extraction methods have been shown to lead to different MCa values (Rosa et al. 2014). Finally, the age difference between participants across studies is non-negligible; the median age for the participants in Syczewska and Święcicka (2016) study is four

years old (Syczewska and Święcicka 2016) compared to 12 years old in the study of Berger et al. (1982) (W. Berger, Quintern, and Dietz 1982). Indeed, we can suspect a significant difference in motor development and gait experiences between the CP groups of these two studies, which may affect the reported MCa values. Thus, a general conclusion regarding potential MCa differences between hemiplegic and diplegic individuals cannot be drawn due to the inconsistency of results and lack of CP-type comparison.

3.6.5.2 Affected vs. non-affected leg

Only one study reported the effect of gait speed on MCa, which is important to take into consideration when investigating hemiplegic gait patterns. The excessive MCa on the affected leg when speed is increased has been related to the altered velocity threshold of the stretch reflex due to the brain lesion (Gross et al. 2013). Vinti et al. (2018) also confirmed that hypothesis by reporting higher MCa in the affected compared to the non-affected leg around the ankle joint (M. Vinti et al. 2018). However, even if Maltais et al. (2004) reported greater MCa around the knee joint, they did not conclude the increased MCa around the ankle (Maltais et al. 2004). Against expectations, Keefer et al. (2004) reported similar or higher MCa on the non-affected compared to the affected leg around the knee (Keefer et al. 2004). It has been suggested that greater MCa on a non-affected leg might represent a limb compensatory adaptation (i.e., increased joint stability to cope with altered motor control). Thus, the literature suggests that excessive MCa on the non-affected leg could be a compensatory mechanism, whereas excessive MCa on the affected leg might result from a cerebral lesion such as spasticity.

3.6.5.3 Shank vs. thigh

Based on the somatotopic arrangements of the corticospinal pathways (i.e., each part of the motor cortex corresponds to a different part of the body), the motor control of distal joints is lower than that of proximal joints. Thus, the greater MCa around the ankle compared to MCa around the knee, reported by Unnithan et al. (1996) (Unnithan, Dowling, Frost, and Bar-Or 1996; Unnithan, Dowling, Frost, Volpe Ayub, et al. 1996), is expectable; however, Leonard et al. (1991) (Leonard, Hirschfeld, and Forssberg 1991) and Maltais et al. (2004) (Maltais et al. 2004) reported higher MCa around the knee compared to the MCa around the ankle. This inconsistency could be

due to the muscles investigated as Maltais et al. (2004) (Maltais et al. 2004) studied the activity of the global quadriceps muscle group (including RF), whereas Unnithan et al. (1996) (Unnithan, Dowling, Frost, and Bar-Or 1996; Unnithan, Dowling, Frost, Volpe Ayub, et al. 1996) only monitored VL muscle. Potentially, higher MCa could be driven by biarticular muscles due to the more complex movement role. According to these findings, more research is needed to conclude the difference between MCa around the ankle and around the knee in individuals with CP.

3.6.5.4 MCa & O_2 cost of walking

Only three studies (D. L. Damiano et al. 2000; Keefer et al. 2004; Unnithan, Dowling, Frost, and Bar-Or 1996) investigated the relationship between MCa and energetic cost. With respect to the O_2 cost of walking, the effect of MCa is unclear: Excessive MCa has been related to higher O_2 cost of walking (Unnithan, Dowling, Frost, and Bar-Or 1996) and higher gait efficiency (i.e., EEI) (D. L. Damiano et al. 2000). Considering that EEI integrates parameters such as the mean velocity and the relative energy expenditure compared to steady-state heart rate during rest, the contribution of MCa to the O_2 cost of walking cannot be understood from such an index; however, Keefer et al. (2004), found no significant relationship between MCa and the O_2 cost or EEI of walking (Keefer et al. 2004). These inconsistencies may be attributable to the different examination settings (i.e., treadmill (Keefer et al. 2004; Unnithan, Dowling, Frost, and Bar-Or 1996) versus overground walking (D. L. Damiano et al. 2000)), or CP type (i.e., solely hemiplegic CP (Keefer et al. 2004) versus both hemiplegic and diplegic CP (D. L. Damiano et al. 2000; Unnithan, Dowling, Frost, and Bar-Or 1996)).

3.6.5.5 MCa & Gait speed

All three studies that investigate the effect of gait speed on MCa (D. L. Damiano et al. 2000; Gross et al. 2013; Unnithan, Dowling, Frost, Volpe Ayub, et al. 1996) showed consistent results with regard to increased MCa when gait speed is increased. It is known that the affection of the upper motor neuron causes speed-dependent hyperexcitability of the strengthening reflex (i.e., spasticity) (Pandyan et al. 2005). Thus, it is expected that this reflex may also increase when gait speed increases and causes greater MCa. Also, the increased MCa with speed reported for the non-affected limb may be due to compensatory mechanisms (Gross et al. 2013).

3.6.5.6 MCa & joint motion

Only one study investigated the relationship between MCa and the corresponding joint excursions during gait (Gross et al. 2015). The negative relationship between MCa and joint movements highlights the role of MCa in controlling joint motion during gait. That is, MCa of the antagonist muscle opposes the agonist to restrain the movement; however, the lack of relationship that has been reported between MCa around the knee and knee flexion from late stance to initial swing supposed that MCa around the knee is not the cause of knee stiffness in hemiplegic CP. Also, the positive relationship between MCa around the ankle and ankle dorsiflexion in the swing phase suggests that MCa around the ankle is related to spastic drop foot in hemiplegic CP (Gross et al. 2015). Based on this study, the contribution of gait MCa on the active range of motion cannot be stated.

Higher MCa has been related to a lower Gillette Gait Index, which means that gait kinematics are closer to the normal pattern (Syczewska and Święcicka 2016). This study also reported no significant relationship between kinematic deviations and altered MCa, which limits information about the contribution of MCa to gait abnormalities. Future research is needed in this domain since the reduction of gait pathology is one of the main functional objectives of CP rehabilitation plans. Many other associations must be investigated to understand better the contribution of MCa on locomotion, such as its effect on gait stability and joint coordination. Indeed, the investigation of abnormal MCa on inter-joint coordination may contribute to a better understanding of the role of MCa on the gait strategy (i.e., coordination pattern) used to walk with stability successfully (Dussault-Picard et al. 2022).

3.6.6 Limitations

The current study has some limitations. First, we were not able to compare MCa in the CP group with that of a healthy group in all studies. In addition, studies included various CP types (e.g., hemiplegia and diplegia) and GMFCS levels (e.g., I and II), and this scoping review does not allow to dissociate MCa between the classifications. Also, EMG experimental protocols and signal analysis were inconsistent: not all studies followed SENIAM recommendations nor stated whether they had normalized EMG prior to MCa computation or not. Different approaches were

implemented for studies that reported their EMG normalization method (e.g., peak vs mean EMG value normalization), limiting comparison across studies. One of the main limitations revealed in this scoping review was the multiple MCa calculation methods implemented, making it difficult to compare and summarize MCa patterns across studies. Finally, the wide range of gait protocols used (e.g., walking environment, walking speed, footwear condition, and using assistive devices) may influence the reported MCa and, thus, limits the studies' comparison, especially comparison between different phases of gait.

3.6.7 Recommendations

As Ikeda et al. (1998) explained previously and based on our findings regarding various MCa calculation methods conducted by the studies using only EMG activity, we recommend using the term MCa rather that MCo when quantifying agonist and antagonist simultaneous contraction based on EMG activation. When estimating the muscle moment and quantifying the percentage of the net moment that the antagonist is negating, then the term MCo should be used to express the simultaneous contraction of the agonist and antagonist muscle pair (Ikeda et al. 1998). The use of standardized terminology will facilitate interpretation and communication in the research and clinical field. Moreover, future studies should focus on MCa comparison between CP types (e.g., hemiplegia, diplegia) and GMFCS levels (e.g., I and II). Indeed, eight studies lack mentioning the GMFCS level of their participants, which leads to a genuine gap in the literature concerning how the gross motor function and MCa during gait are related. This information may contribute to a better understanding of MCa during gait by considering the heterogeneity of the population with CP.

One study included in this scoping review compared MCa using normalized and non-normalized EMG (Gagnat, Brændvik, and Roeleveld 2020). The effect of the EMG normalization approach on muscle activity and MCa values appears to be considerable. As it is impossible to recommend the best approach for EMG normalization when computing MCa, using both methods (normalized and non-normalized) in a single study may be reasonable and benefit future research. Also, future research should focus on implementing standard EMG acquisition and processing methods (see

SENIAM recommendations (Welcome to SENIAM n.d.)), which impacts the MCa validity. Furthermore, it is not possible to determine the best MCa computation method.

Nonetheless, the choice of the MCa calculation method should be justified and explicitly stated in research studies. Finally, the current literature lacks information regarding the contribution of MCa on gait deviations (kinematic and kinetic impairments) at specific gait phases. Indeed, since this scoping review highlights the significant contributors of MCa during gait (e.g., spasticity, motor control, and motor selectivity), future research should focus on its contribution to gait pathology to guide better gait management.

3.7 Conclusion

This scoping review summarized MCa patterns of individuals with CP and their healthy peers during gait across 21 studies. According to existing research, MCa appears to increase in individuals with CP, compared to healthy peers, and it is mainly attributable to cerebral lesion repercussions such as spasticity, altered motor control, and loss of selectivity; however, some inconsistencies between studies make the understanding of MCa difficult concerning, for instance, the influence of the extraction method on the reported MCa value, the MCa differences between different CP types (e.g., hemiplegic, diplegic) and classifications (e.g., GMFCS I and II), and the contribution of the MCa on the cost of walking and gait efficiency.

Chapter 4 – [Pilot experimental study: Muscle co-activation in children with cerebral palsy during gait]

This project was presented at the Orthopaedic Research Society in February 2022 as a poster, and a complete manuscript, comprised of additional data, is in preparation. The research was done with the collaboration of Sahar Mohammadyari Gharehbolagh, Vishnu Deep Chandran, Saikat Pal, and Philippe Dixon. Sahar Mohammadyari Gharehbolagh was involved in the data analysis, interpretation of the results, and providing the abstracts and posters. Vishnu Deep Chandran contributed to the data analysis steps and coding. Saikat Pal helped provide feedback on the abstract drafts. Philippe Dixon was responsible for leading the project, providing the database, and reviewing the abstracts and posters. The following sections present a more in-depth version of the Orthopaedic Research Society abstract.

4.1 Introduction

CP occurs early in life, in an immature brain, posing many problems in the musculoskeletal and motor control systems (Agarwal and Verma 2012). The risk of CP occurs mainly during conception, pregnancy, the perinatal period, and up to the age of two (McIntyre et al. 2011), which means that great attention should be paid to the management of CP during younger ages. Recently, a meta-analysis reported that the prevalence of CP is 1.6 per 1000 in high-income countries over 41 regions of 27 countries classified as low-, middle-, and high-income (McIntyre et al. 2022).

Most children with CP have delayed gait, often with assistance (e.g., upper limbs or walkers) (Miller 2020). The forces of growth and development interact with neural deficits leading to an abnormal gait pattern in children with CP. Moreover, in children with CP, motor system impairments can result in abnormal muscle activity, negatively influencing walking ability. In children with CP, the musculoskeletal system adapts to atypical control, such as spasticity and increased joint stiffness affecting the gait (Miller 2020). Also, these children have shorter and smaller muscle fibres with decreased diameter compared to TD peers (J. Rose et al. 1994).

Muscle structure, activation pattern and co-activation affect CP gait. As 75% of children with CP are ambulatory (Hutton, Colver, and Mackie 2000) and due to the importance of walking tasks in daily life activities (Chiou and Burnett 1985), it is important to enhance the gait efficacy in this population. One component of gait patterns in children with CP is an abnormal level of MCa in lower limb muscles. MCa is the simultaneous activation of agonist and antagonist muscles crossing a joint. Although MCa can occur in normal gait, investigating its pattern in children with CP, regardless of its etiology, might help gait rehabilitation. Therefore, comparing lower limb MCa during walking in children with CP with normative values seen in TD peers is needed.

Following our findings from the scoping review, we identified our research questions for an experimental study. The scoping review demonstrated that MCa is increased in individuals with CP during gait. We aimed to examine this finding in a population including only children with CP. Accordingly, the research question was 1) are MCa around the ankle and knee are greater in children with CP, compared to TD children, across different gait phases (i.e., entire stride, stance, and swing)? Next, our scoping review revealed inconsistencies in methodologies such as EMG processing and MCa calculations across the literature. In particular, there was no consensus on the EMG normalization approach, recommending future research to examine the effect of the normalization of EMG on the resulting MCa values. Therefore, we designed the related research question as 2) is there any difference between CP and TD groups in MCa using normalized and non-normalized EMG? Finally, the literature lacked the comparison of MCa between various CP classifications (i.e., Gross Motor Function Classification System (GMFCS)), and we addressed it in our study by answering this question 3) is there any difference in MCa between children with GMFCS levels I and II?

Thus, the hypotheses were: 1) MCa is greater in children with CP than in the TD group around both ankle and knee joints during all gait phases, 2) EMG normalization affects the MCa results of both muscle groups in both CP and TD groups during the entire stride, stance, and swing, and 3) MCa is higher in children with GMFCS level II than the groups with level I, for both muscle groups during the entire stride, stance, and swing.

4.2 Method

4.2.1 Participants

Data from twelve children with Diplegic CP (4 females, 8 males) and twenty-three TD children (11 females, 12 males) were extracted from a laboratory database (Table 4) (P. Dixon 2015). GMFCS classifications for the CP group were 7 (I) and 5 (II). The age range of participants was between six and eighteen. We extracted the anthropometric characteristics (e.g., sex and age) from the original dataset. Also, EMG data from both right and left legs in the CP group, and for the TD group, either from one leg or both legs, were included; however, we only considered the left side for the current study due to the better quality of the EMG signals for both groups. In the TD group, the exclusion criteria were any history of gait or musculoskeletal abnormality. For the CP group, inclusion criteria were diagnosis of spastic diplegic CP, level of I or II, and independent walking ability. The CP group's main interventions were hamstrings, gastrocnemius Botulinum Toxin injections, and serial casting. CP groups had gait patterns classified as True equinus, Jump, Crouch, mild, and not specified. The local ethics committee approved the original study, and all children provided written assent/consent before participation for inclusion in further research (P. Dixon 2015).

Table 4 - Anthropometric characteristics of the groups

Anthropometric	Groups	
	CP (n=12); mean/median [CI]	TD (n=23); mean [CI]
Age (years)	12.75 [11.03,14.46]	11.04 [9.66,12.42]
Height (m)	1.5 [1.4,1.6]	1.5 [1.4,1.5]
Body mass (kg/m^2)	Median : 39.41 (32.85,45.98)	44.25 [37.43,51.08]

Abbreviations: CP: cerebral palsy, TD: typically developing, CI: confidence interval.

4.2.2 Experiment

In the original study, Oxford Gait Laboratory protocols were followed for the preparation and data collection (P. Dixon 2015). Participants were fit with the skin-mounted retro-reflective markers as per the Oxford Foot Model (Stebbins et al. 2006), and lower-limb Plug-in Gait marker sets were used (Kadaba, Ramakrishnan, and Wootten 1990). A 12-channel wireless system (Wave, Cometa Srl., Milan, Italy) recorded the muscle activity from the following muscles, Rectus Femoris (RF), Semitendinosus (ST), Lateral Gastrocnemius (LG), and Tibialis Anterior (TA), using surface EMG electrodes, sampled at 1000 Hz frequency. Walking trials included data from self-selected, barefoot walking on a 10.0-meter straight walkway.

4.2.3 EMG and gait measurements

Data were imported into Matlab (R2020a, The Mathworks, Inc. Natick, USA), where the open-source biomechZoo Toolbox (v1.8.2) (P. C. Dixon et al. 2017) was used to process and visualize the EMG data. Concerning the EMG signal processing of raw data, we followed these steps: 1) Butterworth bandpass filter (20–500 Hz, fourth order), 2) full-wave rectification, and 3) calculation of RMS to capture the EMG envelope. In another step, the root mean square (RMS) values of EMG signal were dynamically normalized to the peak value found within each trial for each muscle. We did time normalization to 100% of the gait cycle only after calculating MCa to graph the results. In addition, according to the marker coordinate algorithm introduced by Zeni et al. (2008), kinematic gait events were identified (i.e., left foot strike and left foot-off), and then events were used to partition the data (Zeni, Richards, and Higginson 2008). We partitioned the

data into one gait cycle from the first left foot strike to the second left foot strike and identified stance (i.e., first left foot strike to left foot off) and swing (i.e., left foot off to second left foot strike)

4.2.4 Muscle co-activation analysis

We calculated MCa for the thigh and shank muscles, RF/ST, and TA/LG, respectively. MCa was calculated using the method described by Winter (2005) (David A. Winter 2005). This approach is based on the ratio of the common area under the EMG curves of a muscle pair to the sum of the areas under each muscle in that pair (Equation 2.2). MCa values from individual walking trials were averaged for each participant. We calculated MCa from the entire stride and two phases of stance and swing. The total of 38 gait cycles for the CP group and 58 gait cycles for the TD group were evaluated. In order to evaluate the effect of normalization, both non-normalized and normalized EMG signals were included in separate analyses. A representation of MCa calculation for one child with CP is shown in Figure 6.

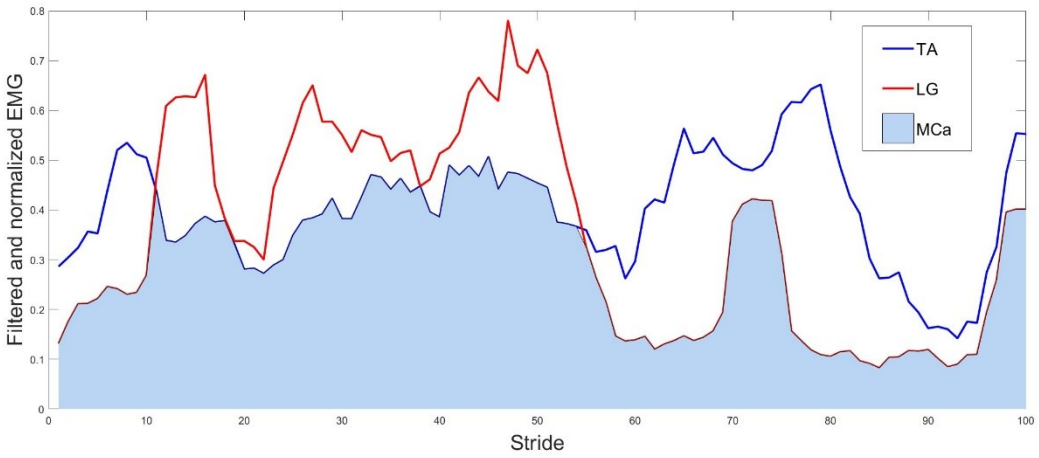


Figure 6. – Sample MCa measurement using normalized EMG for a single child with CP during an entire stride. The blue region is the common area between the Tibialis Anterior (TA) and Lateral Gastrecnemious (LG) muscle pair. The ratio of the blue region to the sum of the areas under each muscle represents MCa.

4.2.5 Statistics

We compared the average MCa from the CP and TD groups for the entire stride, stance, and swing phases using both normalized and non-normalized EMG signals. Statistical analyses were carried out in Matlab. Normality was assessed using Lilliefor's test, and Levene's test was used to assess the equality of variances between groups. If the data were normally distributed, an unpaired t-test (i.e., calculating the group mean) was implemented, otherwise, we used a signed-rank test (i.e., calculating the group median) to compare the groups. For all anthropometric data and MCa, the mean for each group with a 95% confidence interval were calculated and displayed. The significance level was set at $p < 0.05$.

4.3 Results

4.3.1 MCa in CP Vs TD

No significant difference between CP and TD groups was found for age (mean 11.6 ± 3.0 years) ($p = 0.12$), height (mean 1.5 ± 0.2 meters) ($p = 0.97$), and body mass (median (CI) = 42.6 (37.7, 47.4) kg/m^2) ($p = 0.21$). Results from MCa comparison using non-normalized EMG signal analyses showed that for both muscle pairs, MCa was higher ($p < 0.001$) in the CP group than TD during the stride, stance, and swing (Table 5). MCa measurement using normalized EMG showed different results (Table 6). There was no significant difference between groups for MCa of both (RF/ST) and (TA/LG) muscle pairs during the entire stride ($p = 0.08$ and $p= 0.09$, respectively). During stance, the difference between groups was insignificant for the thigh ($p = 0.11$), whereas it was significantly higher in CP group for the shank muscles ($p < 0.001$). MCa around the ankle and knee were significantly higher ($p=0.03$ and $p < 0.001$, respectively) in CP group than TD during the swing.

Table 5 - MCa results calculated from non-normalized EMG for both CP and TD groups

Gait Phases	Thigh (RF/ST) (mean [CI]; p < 0.001)		Shank (TA/LG) (mean/median [CI]; p < 0.001)	
	CP	TD	CP	TD
Stride	61.08 [56.39,65.76]	45.03 [42.63,47.44]	Median: 67.70 (62.49,72.91)	51.13 [48.52,53.74]
Stance	46.59 [42.03,51.15]	34.19 [30.90,37.48]	59.73 [55.50,63.96]	43.84 [41.50,46.19]
Swing	66.76 [62.90,70.62]	51.39 [48.54,54.25]	Median: 47.21 (41.50,52.91)	29.99 [28.18,31.79]

Abbreviations: CP: cerebral palsy, TD: typically developing, RF: Rectus Femoris, ST: Semitendinosus, LG: Lateral Gastrocnemius, TA: Tibialis Anterior, CI: Confidence Interval.

Table 6 - MCa results calculated from normalized EMG for both CP and TD groups

Gait Phases	Thigh (RF/ST) (mean/median [CI])		Shank (TA/LG); (mean/median [CI])	
	CP	TD	CP	TD
Stride	55.48 [51.84,59.11]	51.149 [48.046,54.252]	59.26[55.08,63.44]	54.69 [51.35,58.03]
Stance	51.83 [46.06,57.59]	Median: 46.25 (41.72,50.79)	*54.91 [51.48,58.33]	43.29 [39.87,46.71]
Swing	*59.33 [55.78,62.89]	51.65 [47.35,55.96]	*51.18 [45.78,56.58]	Median: 33.39 (29.59,37.19)

Abbreviations: CP: cerebral palsy, TD: typically developing, RF: Rectus Femoris, ST: Semitendinosus, LG: Lateral Gastrocnemius, TA: Tibialis Anterior, CI: Confidence Interval, *: significant.

4.3.2 MCa in CP Classifications

No significant differences between children with GMFCS I (n=7) and II (n=5) were found for age (mean 12.75 ± 2.8 years) (p = 0.64), height (mean 1.5 ± 0.1 m) (p = 0.95), and body mass (median (CI) = 39.41 (32.85, 45.98) kg/m2) (p = 0.96). We compared MCa between groups using both normalized and non-normalized EMG. Regardless of EMG normalization, MCa values were similar in both groups for thigh (RF/ST) and shank (TA/LG) muscles across the entire stride and stance phase. During the swing, however, MCa around the knee calculated from normalized EMG was greater in GMFCS (II) compared to GMFCS (I) (median CI = 63.87 (58.70,69.04) and mean CI = 56.22 [51.57,60.88], respectively, p = 0.019), and no significant difference was seen for MCa

calculated from non-normalized EMG. Groups were similar for MCa around the ankle during the swing for both normalized and non-normalized EMG ($p<0.05$).

4.4 Discussion

This study aimed to investigate if there were differences in lower-limb MCa between children with CP and their TD peers during walking (i.e., stride, stance, and swing), as well as between children with GMFCS I and II. Also, we aimed to identify the effect of the EMG normalization on the MCa values. Now we discuss our main findings based on our hypotheses.

According to the results, our first hypothesis was confirmed for MCa values using non-normalized EMG, whereas it was partially confirmed for MCa values using normalized EMG. The difference in results showed that EMG normalization impacts MCa values, confirming our second hypothesis. The lack of significant difference in MCa using normalized EMG between groups during the stride highlights the effect of the EMG normalization on removing the variability between individuals (discussed in chapter 5). On the other hand, comparing MCa across phases (i.e., stance and swing) appears to lessen the effect of normalization, as it was shown that MCa around the ankle is significantly higher in CP compared to TD when comparing during stance and swing. An Evaluation of MCa during more sub-phases (e.g., initial stance, mid-stance) in future research could clarify this argument. Also, the impact of EMG normalization appeared insignificant when comparing MCa within CP (i.e., GMFCS I and II), as the difference between groups was only seen for MCa around the knee during the swing. This might be due to the small sample size in our study (i.e., 7 vs 5).

Significant differences between groups seen during the stance and swing emphasized the necessity of evaluating MCa across various gait phases and sub-phases. For instance, in our study, MCa around the ankle and knee (normalized EMG) significantly differed between CP and TD groups during the swing, whereas, during stance, the difference was seen only around the ankle. The importance of the independent evaluation of MCa during the stance and swing phases in gait studies has been stated before (Lo et al. 2017), and it would be helpful to consider kinetic and kinematic adaptations when comparing MCa during various gait phases. A possible explanation for greater MCa around the ankle is that children with CP maintain their stability around the ankle

joint mostly through contracting the surrounding muscles, especially to compensate for the lack of plantar flexor muscle power (Schweizer, Romkes, and Brunner 2013). Shank muscles play a key role in abnormal gait patterns, particularly their distal part, which is more sensitive to injuries and neurological insults, and their inefficient activity affects both the ankle and knee joints. Furthermore, children with CP may have relied on an ankle-dominant strategy for balance control during gait (Rethwilm et al. 2021), leading to a greater MCa in the shank muscles.

To our knowledge, only one study compared MCa values between CP and TD groups during walking using both normalized and non-normalized EMG (Gagnat, Brændvik, and Roeleveld 2020). A direct comparison between our study and Gagnat et al. (2020) is not possible as they evaluated MCa in various gait sub-phases (e.g., weight acceptance, mid-stance). However, some consistencies can be interpreted, such as similar MCa values around the knee joint between CP and TD groups when using normalized EMG during the initial stance. In other words, both studies revealed an insignificant difference in MCa values around the knee using normalized EMG between CP and TD groups. On the other hand, our results regarding MCa around the knee using non-normalized EMG were not consistent with Gagnat et al. (2020) (i.e., despite us, they found no significant difference between CP and TD). Differences between studies might be due to various MCa calculation methods and various CP types (i.e., diplegic and hemiplegic) included by Gagnet et al. (2020).

Our third hypothesis was partially confirmed. To our knowledge, this is the first study comparing MCa between different GMFCS classifications. Our results showed no significant difference in the MCa values around the ankle and knee during the entire stride and stance phase between GMFCS groups (i.e., I and II). Nonetheless, the results demonstrate that MCa around the knee calculated using normalized EMG was significantly higher in Children with GMFCS II during the swing. MCa during the stance is associated with physical parameters, while during the swing, it is mainly associated with cognitive parameters (Lo et al. 2017). Thus, greater MCa around the knee in children with GMFCS II during the swing may be partly due to the more impaired cognitive abilities and motor activation levels.

This study has some limitations. First, we could not include children with hemiplegic CP to compare MCa between CP types (e.g., Hemiplegic vs diplegic). Furthermore, our study was under power regarding the sample size of GMFCS groups (i.e., 7 vs 5). The opportunity for increased sample size, including various CP types, provides this possibility in future studies. Another potential limitation is that we only considered single muscle pairs in MCa calculations. For MCa around the thigh and ankle we included RF/ST and TA/LG muscle pairs, respectively. Considering more muscles when comparing MCa in dynamic activities such as walking might be more reliable, specifically for the bi-articular muscles like RF and gastrocnemius. Moreover, Muscles contribute differently to Triceps Surae activation (i.e., Gastrocnemius Medialis (GM) (˜ 24%), Soleus (So) (˜ 60%), and LG (˜ 16%)) (Morse et al. 2005). The dependence of the heterogeneity of the muscle activity to a specific muscle, individual, and task has been examined previously. For example, it has been shown that only GM was heterogeneously active during single-leg heel raise (Kinugasa, Kawakami, and Fukunaga 2005). However, another study revealed heterogeneity of all three muscles (i.e., GM, So, and LG) for the same task (Segal and Song 2005). As these studies did not evaluate the walking task, generalizing the findings to all other movements might not be logical. However, making a concrete conclusion about the interpretation of MCa around the ankle only based on one single calf muscle (i.e., LG in our study) seems to be limited.

4.5 Conclusion

Our present study provides new evidence supporting different levels of MCa in children with CP compared to their TD peers during walking for thigh (RF/ST) and shank (TA/LG) muscles. Our study revealed that the normalization of EMG signals, gait phases, and GMFCS levels could influence MCa measurements in children with CP. Future work includes analysis of MCa concerning various CP types, namely, hemiplegic and diplegic, and during more detailed gait phases (i.e., initial stance, mid-stance). A more straightforward walking pattern is known as the primary goal in CP gait rehabilitation, which can be achieved by treating the altered muscle activity and MCa patterns (Novak et al. 2013). Therefore, knowledge of MCa could provide clinicians with insights into gait disorder management and applying proper interventions to these populations' specific needs.

Chapter 5 – [Discussion]

The general objective of this master's project was to compare muscle co-activation (MCa) during gait in children with cerebral palsy (CP) with their typically developing (TD) peers. This objective was achieved through two studies.

First, we conducted a scoping review to better identify muscle co-contraction (MCo)/MCa patterns in individuals with CP during gait and find the knowledge gaps in this domain. The results of this study are presented in chapter 3. This study enabled us to distinguish different terminologies (i.e., MCo and MCa) and then quantify the difference in MCa values during walking between individuals with CP and their healthy peers. Also, we could examine the effect of some of the gait parameters, such as joint motion and speed, on MCa. Moreover, the scoping review revealed inconsistencies between studies regarding gait protocols, electromyography (EMG) analysis, MCa calculation methods and the necessity of comparing CP types and classifications.

Next, we applied the scoping review findings to a cross-sectional observational study that compared MCa during walking in children with CP with their TD peers. The results of this study are presented in chapter 4. Comparison between CP and TD groups revealed that the normalization of EMG signals alters MCa values. Groups were significantly different only during the stance and swing for MCa around the ankle in shank (Tibialis Anterior (TA)/Lateral Gastrecnemious (LG)) muscles and during the swing for MCa around the knee for the thigh (Rectus Femoris (RF)/ Semi-tendinosis (ST)) muscles using normalized EMG. This study also highlighted the importance of comparison of MCa during different phases of gait. Although we compared MCa during only two main gait phases (i.e., stance and swing), differences between groups demonstrated the effect of the various muscular activations happening during specific phases of gait on MCa patterns in children with CP. In addition, children with Gross Motor Function Classification System (GMFCS) II showed greater MCa using normalized EMG around the knee joint during swing compared to children with GMFCS I.

This fifth chapter begins with a discussion of our results, considers the links between them, and highlights their importance in contributing to the existing knowledge. We will first contrast the

terminologies (i.e., MCo and MCa) (section 5.1), followed by a discussion about EMG processing and MCa calculation methods (section 5.2). We will then discuss the results related to the difference in MCa values between CP and TD groups, highlighting the effect of the EMG normalization (section 5.3). Then, we will discuss the comparison of MCa between two GMFCS levels (i.e., I and II) (section 5.4), followed by a highlight of the strengths and limitations of this master's project (section 5.5). The chapter will conclude with the perspectives for future research (section 5.6).

5.1 Co-activation vs. co-contraction

One of the considerable ambiguities across the literature was the different terminologies used by authors while reporting on the simultaneous activation of agonist and antagonist muscles crossing the same joint. We addressed this issue in our scoping review. According to our findings, a distinction should be made between the terms MCa and MCo. Ikeda et al. (1998) attribute different terminologies to different methods for calculating the simultaneous activation of agonist and antagonist muscle pair (i.e., MCa when using EMG and MCo when using muscles moment). Another study, however, only highlighted different methodologies and not terminologies (i.e., MCo can be calculated using both EMG and moment) (Knarr, Zeni, and Higginson 2012). It is known that greater EMG magnitude is related to greater muscle force crossing a joint (Richards and Higginson 2010; Tm and F 2005). However, the relationship between EMG activity and muscle force is not linear (Thomas S. Buchanan et al. 2004). Although it is possible to calculate MCo using EMG-to-force models (Knarr, Zeni, and Higginson 2012; Souissi et al. 2017), this method is time-consuming and difficult in clinical research. The difficulty is mainly due to the complex data collection, critical electrode placement, and normalization methods associated with these models (Knarr, Zeni, and Higginson 2012). Furthermore, although using EMG-based methods does not distinguish the force production capacity of the agonist and antagonist muscles, they are more recommended in clinical settings (Chandran et al. 2019). Therefore, despite existing studies using MCa and MCo interchangeably, no matter which method was implemented (i.e., EMG vs moment), we followed the recommendations of Ikeda et al. (1998) and expressed simultaneous EMG activation of the agonist and antagonist muscles crossing a joint as MCa in our experimental study (Ikeda et al. 1998). It should be noted that the use of specialized

and technical terminologies will simplify the interpretation in the scientific field and helps knowledge advancement.

5.2 EMG processing and MCa calculation method

5.2.1 EMG processing

We summarized EMG acquisition and processing across all studies included in the scoping review (Table 1). Studies were not consistent in EMG analysis methods before MCa measurement. This was in line with the results of previous research on the methodologies for EMG processing across studies evaluating MCa during gait in pathological populations (Rosa et al. 2014). How we process EMG signals affects the results and group comparison. For instance, the intra-individual variability of gait EMG is likely to decrease when smoothing the signal to a great degree (i.e., by increasing the moving average window or a lower cut-off frequency) (A. M. Burden, Trew, and Baltzopoulos 2003). This will lead to similar EMG patterns between groups, which is problematic when comparing healthy groups with neurologic populations such as CP. Therefore, adherence to Surface Electromyography for the Non-Invasive Assessment Muscles (SENIAM) and the International Society of Electrophysiology and Kinesiology standards plays a vital role in the reliability and validity of results in EMG-based studies. We followed the expected standards in EMG processing (Section 4.2.3). For instance, we filtered EMG signals using a fourth-order Butterworth bandpass filter (20–500 Hz) (Standards-for-Reporting-EMG-Data.pdf n.d.).

Another factor worth considering is EMG normalization. Our scoping review presented a range of EMG normalization methods (e.g., to the mean and maximum values recorded during the gait trials, and to the maximum voluntary activation recorded during a maximum voluntary contraction (MVC)). Although normalizing to MVC is known as the most popular method, it should only be used in studies with healthy and trained subjects (Konrad 2005). It is likely that MVC tests easily get invalid and produce invalid data (Konrad 2005), and it is less likely to be representative of the maximum capacity of the muscle in individuals with neurological problems (A. Burden and Bartlett 1999). In individuals with CP, for example, it may overemphasize the contributions of weaker, more spastic, or less effective muscles. Instead, the mean value and maximum value

reached within a period are known to be feasible methods for EMG signals normalization from the neurological population (Yang and Winter 1984).

Our scoping review recommended that if normalizing the EMG signals before computing the MCa, no matter to which reference value, it is helpful to compare the results with MCa calculated using non-normalized EMG. Also, it has been suggested not to normalize EMG data in individuals with neurological impairments (Gagnat, Brændvik, and Roeleveld 2020). Other researchers confirm this idea by stating that because most MCa calculation approaches are based on a ratio, non-normalized EMG prevents unnecessary data transformation (Banks et al. 2017). Furthermore, EMG values from weak muscles are usually low during walking, and using normalized signals (e.g., to the percent of a maximum value) may exaggerate the MCa values (Lamontagne, Richards, and Malouin 2000). Thus, we decided to consider both methodologies (i.e., using normalized and non-normalized EMG) in our experimental study.

5.2.2 MCa calculation method

Our scoping review presented a range of MCa calculation approaches. Although a few approaches were more used, their validity remained unclear in studies including the population with CP. Rosa et al. (2014) did not recommend any particular methodology to assess MCa. They included all the literature studying MCa during gait in people with neurological impairments, including CP (Rosa et al. 2014). Therefore, in our experimental study, we decided to calculate MCa using a well-established method introduced by Winter (2005) as the percentage of total muscle activity when agonist and antagonist muscle pair (TA/LG) and (RF/ST) were simultaneously activated. In this method, a co-activation of 100% does not mean that ‘‘there will be no movement” but means that both muscles had simultaneous activation at the same intensity and time (Candotti et al. 2009). Only one study included in the scoping review implemented this method; however, the study did not compare results in children with CP with a healthy group (Keefer et al. 2004).

5.3 MCa in children with CP vs. TD peers

Our scoping review revealed increased MCa values in individuals with CP, compared to healthy peers during walking for various thigh and shank muscle pairs. We examined this result in our

experimental study as our first research question, comparing two groups of children with CP and TD. Moreover, we compared MCa between the groups using both normalized and non-normalized EMG activity of the muscle pairs. In the following sections, the results related to each approach are discussed.

Among those studies in the scoping review reporting whether or not they normalized EMG, none used non-normalized EMG solely to compare MCa between CP and healthy groups (Table 1). Only one study (Gagnat, Brændvik, and Roeleveld 2020) included both normalized and non-normalized EMG, and we discussed their results in the previous chapter. We decided to include non-normalized EMG in our analysis in order to investigate the effect of the EMG normalization on the resulting MCa as recommended by the scoping review. Following this analysis, children with CP demonstrated greater MCa around the knee and ankle joints during the entire stride, stance, and swing phases compared to TD children. Our finding is consistent with the studies by Berger et al. (1982) and Leonard et al. (1991), reporting greater MCa around the ankle (i.e., TA/GM and TA/LG, respectively) and knee (i.e., RF/BF for both studies) during gait in CP compared to the healthy group; however, we are not aware if they normalized EMG or not (Table 2) (W. Berger, Quintern, and Dietz 1982; Leonard, Hirschfeld, and Forssberg 1991). Due to lack of evidence, comparison between studies is limited. This highlights the necessity of reporting more details about the EMG analysis steps in MCa studies to contribute to knowledge transfer.

Our results confirmed the impact of the normalization on the muscle activity and co-activation explained before, emphasizing the importance of considering both non-normalized and normalized EMG when calculating MCa. Despite the findings from non-normalized EMG, the difference between groups during the entire stride for both muscle pairs and during the stance for MCa around the knee was insignificant when using normalized EMG. This demonstrates that the normalization process has decreased the variability between children compared to using non-normalized EMG. Previous research revealed that peak dynamic normalization decreases the variability of the gait EMG of individuals compared to non-normalized EMG (Halaki et al. 2012). Furthermore, this normalization method increases the homogeneity by eliminating the biological differences within the group more than other normalization methods, such as normalizing to MVC

(Allison, Marshall, and Singer 1993; Knutson et al. 1994). This improved homogeneity might not be desirable in studies including pathological populations such as CP.

Nonetheless, using normalized EMG, the CP group displayed greater MCa around the ankle compared to TD peers during the stance and greater MCa around the knee and ankle during the swing. Our findings are partially consistent with the study by Unnithan et al. (1996). Although they normalized EMG to the maximum value, they reported greater MCa values in children with CP compared to the healthy group for both shank (TA/So) and thigh (Vastus Lateral (VL)/ Medial Hamstring (MH)) (Unnithan, Dowling, Frost, Volpe Ayub, et al. 1996). According to Wakeling et al. (2007), thigh muscles (RF/ Semi-membranosus (SM) had relatively normal function compared to the shank (TA/Gastrocnemius Medialis (GM)), (Wakeling, Delaney, and Dudkiewicz 2007) which might explain relatively similar MCa around the knee during the stance between CP and TD groups. However, greater MCa around the knee in CP group during the swing might be due to the excessive activation of RF trying to lift the leg and then decelerate the leg, as described in chapter 2.

According to clinical observations, muscle weakness is more pronounced in more distal parts of the limb (Wiley and Damiano 1998), and co-activation is known as a strategy that children with CP adopt due to muscle weakness, especially around the ankle joint (Lorentzen et al. 2019). Greater MCa around the ankle during both the stance and swing phases in our study can be explained by the pronounced muscle dysfunction in this area introduced by Wakeling et al. (2007). Also, it can be attributed to their need to generate power around the ankle during push-off, contributing to the forefoot fulcrum (Basmajian 1962). Moreover, the role of the ankle dorsi flexor (TA) in foot clearance during swing (Wakeling, Delaney, and Dudkiewicz 2007), which leads to an increase in antagonistic muscle activity, should be taken into consideration. Muscles surrounding the ankle joint are known as the main contributors to the abnormal gait pattern, leading to abnormal activity at both ankle and knee joints (Stewart and Shortland 2010). A particular focus should be placed on these muscles in treatment plans.

5.4 MCa within children with CP

Following the recommendations of our scoping review, we compared MCa around the knee and ankle joints during gait between children with GMFCS I and II. Groups were different only for MCa around the knee using normalized EMG during the swing (i.e., higher for GMFCS II). The lack of difference in MCa between the two GMFCS classifications supports previous findings on the lack of a relationship between co-activation and level of function as measured by Gross Motor Function Measure (GMFM) in the study by Damiano et al. (2000). They examined MCa around the knee for (RF/BF) muscle pair in a group of children with CP with GMFM I, II, and III during gait, and reported that children with greater neurologic involvement did not have more co-activation (D. L. Damiano et al. 2000).

In our study, the gait pattern of only two children with GMFCS level II was specified: true equines and mild (i.e., within one standard deviation of typical patterns). Gait patterns for children with GMFCS level I were mild (n=3), jump (n=1), and crouch (n=1). There was not sufficient information about the gait patterns of our subjects, making the comparison and interpretation limited.

5.5 Strengths and limitations

In general, the objective of this master's project was to study MCa around the knee and ankle joints during walking in children with CP and compare it with that of TD peers. Two studies were conducted to achieve this objective: (1) a scoping review of available literature and (2) a cross-sectional observational study. Some strengths and limitations are inherent within each study.

To our knowledge, our submitted scoping review is the first study summarizing all the existing literature on MCa and MCo in individuals with CP during gait. The effect of gait parameters on MCa in individuals with CP and comparisons between CP and healthy groups have been discussed in chapter 3. Despite the wide range of differences across studies included, our scoping review revealed the typical pattern of MCa in individuals with CP and the most pronounced knowledge gaps in this area. Our study highlighted the importance of using the appropriate terminology (i.e., co-contraction vs co-activation). As previous research clearly indicated, when quantifying co-activation based on EMG activity alone, it is MCa being calculated. In contrast, when developing

a biomechanical model to predict individuals' moments produced by agonist and antagonist muscle groups, it is MCo which can be calculated as the percentage of the net moment that is negated by the antagonist moment (Ikeda et al. 1998). In our scoping review, the term MCa was used as muscle moment has been calculated in none of the included studies. Researchers should use the appropriate term based on their specific methodology. Moreover, this study provided researchers with a unique opportunity to investigate MCa considering the knowledge gaps we suggested. As such, one of the main gaps discovered was the importance of considering the effect of EMG processing, namely, normalization on the resulting values and, in turn, the interpretation of findings when comparing the impaired and healthy groups.

Following the conduction of the scoping review, we had the chance to examine some of the research questions raised from this study in a pilot experimental study. We examined two recommendations of the scoping review in this study. First, we compared MCa between children with CP and their TD peers, using normalized and non-normalized EMG activities to interpret the results better. Next, to the best of our knowledge, this is the first study that compared MCa between two GMFCS classifications (i.e., I and II) in children with CP.

Our study has some limitations, which are discussed in the following paragraphs. First, we mentioned some limitations of our scoping review (subsection 3.6.6) in chapter 3, particularly concerning the inconsistencies in gait protocols, EMG experimental protocols, signal analysis, and MCa calculation methods, limiting the comparison across studies and drawing a general conclusion impossible. Consequently, this limited us in choosing our methodologies (i.e., EMG processing and MCa calculation) for the experimental study. Second, according to the literature review (subsection 2.2.3), there are some limitations in measuring MCa using EMG activity. EMG signals can be contaminated by cross-talk from other muscles, such as distant strong antagonist muscles (De Luca and Merletti 1988), or signals from other electrical devices, which was out of our control.

There were some limitations concerning the role of walking performance. One confounding factor was the experience of walking among all children. Because the walking onset is delayed in children with CP, they usually have less walking experience than their TD peers. Apart from that, children

with CP often only walk during the therapy sessions, or more supervised practice time at home, leading to fewer steps per day compared to their TD peers (Bjornson et al. 2007). Thus, they have more muscle activity than children with TD, who are more experienced in walking tasks (Prosser et al. 2010). In general, MCa can be decreased when adaptation to the task is increased. In addition, the effect of gait speed on muscle activity (Schwartz, Rozumalski, and Trost 2008) and MCa for CP and TD groups (Unnithan, Dowling, Frost, Volpe Ayub, et al. 1996) has been studied previously. We discussed the relationship between speed and MCa in our scoping review. The limitation was that children with CP and TD walk at different self-selected speeds (i.e., slower in CP group), affecting their muscle activity and MCa values (Gage et al. 2009). Some authors eliminated this confounding factor. For example, Gross et al. (2013) used a non-dimensional gait speed and a mixed linear model to control inter-subject and intra-subject variability, respectively. They compared groups walking at slow, spontaneous, and fast speeds to prepare for similar conditions and assess MCa on a comparable range of speeds (Gross et al. 2013).

A final limitation is that we were not able to measure MCa from both legs in the CP group (i.e., more affected and less affected legs). Among twelve children with CP, six did not have gait pattern classification on one of their legs, and two had a similar level of impairment in both legs. Therefore, a between-legs comparison was possible only for five participants which had limited power of comparison. We could not address all the knowledge gaps identified from scoping review in our experimental study. For instance, we were limited in choosing our study population who were only diplegic CP (i.e., unable to compare with hemiplegic CP). Moreover, our results might be affected by our small sample size when comparing GMFCS groups (i.e., 7 vs 5). These limitations, nevertheless, offer potential objectives for future research.

5.6 Perspective

This master's project aimed to deliver knowledge about muscular adaptations during walking in individuals with CP, especially children. Particularly, it contributed to understanding muscle co-activation during walking in this population. Some future research perspectives that can further advance this knowledge are discussed.

Increasing the sample size in future research can strengthen the power of results and the ability to generalize them to a larger population. More extensive studies into the MCa in children with hemiplegic and diplegic CP would prepare the possibility of comparisons between CP types. Besides CP type, a comparison between legs (affected vs non-affected, less affected vs more affected) is also recommended in future research. Furthermore, computation of MCa for more detailed gait sub-phases (i.e., initial contact, mid-stance) might be more beneficial for exploring the pathological and adaptive components of MCa in the affected, non-affected, less-affected, and more-affected legs in children with CP and healthy limbs. Finally, a comparison between MCa around the knee and ankle joints for both CP and TD groups would contribute to the knowledge about inter-site differences in MCa.

With regard to methodology, future research might compare groups using different MCa calculation equations and various normalization methods, such as peak and mean dynamic normalization, and normalizing to MVC in a single study. These comparisons evaluate different measurement methods, contributing to the conduction of standard approaches in gait assessment in children with CP.

Chapter 6 – [Conclusion]

The objective of this master's project was to identify the difference in the level of muscle co-activation (MCa) between children with cerebral palsy (CP) and their typically developing (TD) peers at the knee and ankle joints. The two studies presented in two different chapters in this project provided the possibility of gaining a level of knowledge about characteristics of MCa during gait in individuals with CP from the existing literature; to quantify MCa during gait in children with CP and compare it with normative values from a group of TD children.

First, the scoping review conducted on muscle co-contraction (MCo) and MCa in individuals with CP during gait made it possible to understand better the existing knowledge and the missing knowledge across the literature base. The main findings of this review were identifying the difference between terminologies (i.e., MCo and MCa) and generally increased levels of MCa in individuals with CP compared to healthy peers during gait. Nevertheless, the heterogeneity between included studies caused by differences in CP types, gait protocols, EMG analysis procedures, and MCa calculation approach led to some uncertainties about studying MCa in this population. Thus, following standard methodologies for quantifying MCa in a population with CP is required to draw a concrete conclusion about MCa patterns. Furthermore, more studies are needed to address the research gaps in this area to enhance the general interpretation of MCa in individuals with CP.

Then, our experimental study confirmed the effect of the EMG normalization, gait phases, and to some extent, Gross Motor Function Classification System (GMFCS) level of CP on the resulting MCa values. The MCa was increased in CP compared to TD during gait. However, the results were different when normalizing the EMG. Including both normalized and non-normalized EMG in the analysis will help to understand the reason for increased or similar MCa values. Also, the level of functionality affected the MCa values in children with CP considering that this association was changed according to the muscle group and gait phase.

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Appendix 1

Database(s): **Ovid MEDLINE(R) ALL** 1946 to April 08, 2021

Search Strategy:

#	Searches
1	gait disorders, neurologic/ or gait apraxia/ or gait ataxia/
2	gait/ or gait analysis/ or walking speed/
3	locomotion/ or walking/ or dependent ambulation/ or stair climbing/
4	1 or 2 or 3
5	(Gait or walk* or treadmill? or ambulatory or ambulation or locomotion or step or stride or stair? or stairway).ab,kf,ti.
6	4 or 5
7	Cerebral Palsy/
8	("cerebral palsy" or "spastic diplegia" or "bilateral spastic" or "unilateral spastic" or hemiplegia or hemiplegic or "little disease" or "brain palsy" or "brain paralysis" or "central palsy" or "central paralysis" or "cerebral paralysis" or "cerebral paresis" or "encephalopathia infantilis"). ab,kf,ti.
9	7 or 8
10	muscle hypertonia/ or muscle rigidity/ or muscle spasticity/
11	(co-contraction? or cocontraction? or co-activation or synergy or synergies or hypertonia* or spasticity or rigidity or synergistic movement or inter-joint coordination or activation patterns).ab,kf,ti.
12	10 or 11
13	6 and 9 and 12
14	limit 13 to humans
15	limit 14 to (English or French)