



## Pathological and physiological muscle co-activation during active elbow extension in children with unilateral cerebral palsy



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### HIGHLIGHTS

- Muscle co-activation in children with cerebral palsy was pathological and physiological.
- Pathological co-activation increased with elbow extension speed.
- Pathological co-activation was more pronounced in brachioradialis than biceps brachii.

### ABSTRACT

**Objective:** To address the roles and mechanisms of co-activation in two flexor/extensor pairs during elbow extension in children with cerebral palsy (CP).

**Methods:** 13 Typically Developing (TD) and 13 children with unilateral spastic CP performed elbow extension/flexion at different speeds. Elbow angle and velocity were recorded using a 3D motion analysis system. The acceleration and deceleration phases of extension were analyzed. Co-activation of the brachioradialis/triceps and biceps/triceps pairs was computed for each phase from surface electromyographic signals. Statistical analysis involved linear mixed effects models and Spearman rank correlations.

**Results:** During the acceleration phase, there was strong co-activation in both muscle pairs in the children with CP, which increased with speed. Co-activation was weak in the TD children and it was not speed-dependent. During the deceleration phase, co-activation was strong and increased with speed in both groups; co-activation of brachioradialis/triceps was stronger in children with CP, and was negatively correlated with extension range and positively correlated with flexor spasticity.

**Conclusions:** Abnormal patterns of co-activation in children with CP were found throughout the entire movement. Co-activation was specific to the movement phase and to each flexor muscle.

**Significance:** Co-activation in children with CP is both physiological and pathological.

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## 1. Introduction

Cerebral palsy (CP) is a neurological pathology caused by a defect or lesion of the immature brain, which leads to disorders of movement and posture. CP is characterized by a combination of motor impairments, including spasticity, hyper-reflexia, muscle weakness, loss of selective motor control and excessive muscle co-activation (CA). These motor impairments can severely limit activities of daily living (O'Shea, 2008).

**Abbreviations:** AROM, active range of motion; CA, co-activation; CP, cerebral palsy; EAccP, extension acceleration phase; EDecP, extension deceleration phase; EF, extension/flexion; EMG, electromyography; IUL, involved upper limb; MACS, Manual Ability Classification System; MAS, Modified Ashworth Scale; PPV, Percentage to Peak Velocity; SUCP, Spastic Unilateral Cerebral Palsy; TD, Typically Developing.

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Muscle CA is the simultaneous activation of an agonist muscle, which causes movement at a joint in a particular direction, and an antagonist muscle, which opposes the movement (Ikeda et al., 1998). Physiologically, muscle CA induces mechanical co-contraction of muscles, the role of which is to regulate joint stiffness (Bullock and Grossberg, 1991). Appropriate levels of co-contraction are required during sudden changes in the direction of joint motion, fine motor activities, and to stabilize loaded joints (Humphrey and Reed, 1983; Johansson and Westling, 1988; Valero-Cuevas, 2005). Any alteration in the capacity to regulate the level of co-contraction has a direct impact on movement.

It is well established that children with CP often have excessive CA of the muscles of both lower limbs (Leonard et al., 1991; Unnithan et al., 1996; Ikeda et al., 1998; Prosser et al., 2010; Gross et al., 2013, 2015) and both upper limbs (Feltham et al., 2010; Braendvik and Roeleveld, 2012; Sarcher et al., 2015; Xu et al., 2015). Specifically, active elbow extension of the involved upper limb (IUL) in children with spastic unilateral CP (SUCP) induces excessive CA (Van Thiel et al., 2000; Volman et al., 2002; Sarcher et al., 2015) because of excessive activation of the flexor muscles, which are spastic (Sarcher et al., 2015).

However, it is not yet clear whether the effect of this excessive CA is actually negative, i.e. if it restricts movement (henceforth termed pathological CA), or if it also serves to increase joint stability in patients who also have muscle weakness (henceforth termed physiological CA). Some studies have suggested that there is a relationship between excessive CA and reduced peak elbow velocity (Van Thiel et al., 2000; Rameckers et al., 2010), and between excessive CA and restricted elbow active range of motion (AROM) (Levin et al., 2000; Sarcher et al., 2015). However, there are discrepancies in the literature regarding the effect of reducing muscle hyperactivity, including CA, on motor capacity, by the use of intramuscular injections of botulinum toxin type A. Some studies have found improvements in motor capacity after botulinum toxin A injections in the upper limbs (Lee et al., 2013; Ferrari et al., 2014; Sakzewski et al., 2014; Lidman et al., 2015) while others have found little or no improvement due to the concomitant reduction in strength and loss of necessary, functional CA (Fehlings et al., 2000; Rameckers et al., 2007, 2009; Hoare et al., 2010; Olesch et al., 2010; Speth et al., 2015).

The discrepancies between the results of studies investigating the effect of chemodenervation on upper limb movements are likely related to two issues: On one hand, there is a lack of understanding of the CA mechanisms in the IUL of children with SUCP. Van Thiel et al. (2000) hypothesized that excessive CA in the IUL occurs particularly at the end of the extension movement, when the need for joint stability is the greatest. It is thus necessary to carry out separate analyses of CA during the acceleration and deceleration phases of the movement, in order to determine at which point of the movement the pathological CA occurs. To our knowledge, this has not yet been done. Moreover, movement speed has been shown to increase muscle activation and CA levels during gait, more significantly in the lower limb of children with CP than in the lower limb of Typically Developing (TD) children (Gross et al., 2013). Although some studies of upper limb movements have included the notion of speed (Van Thiel et al., 2000; Rameckers et al., 2010) by analyzing “fast” movements, presumably to induce higher levels of CA, to our knowledge the effect of movement speed on CA in the IUL of children with SUCP has never been quantified. A thorough analysis of the movement conditions under which CA in the IUL of children with SUCP differs from CA in the upper limbs of TD children may provide insights into the different roles of CA, both for diagnostic purposes and to optimize treatment effectiveness.

On the other hand, there is little evidence regarding which muscles should be targeted by chemodenervation treatments, such as botulinum type A injections. Indeed, it remains unclear whether

spastic muscle overactivity differs between different muscles that produce similar movements, and the extent to which it interferes with these movements. In order to determine the muscles which are the most affected, it is necessary to analyze the activation of individual elbow flexor muscles during elbow movement. This would enable the development of appropriate guidelines for treatment by botulinum toxin type A injection.

Therefore, the primary aim of this study was to highlight the different roles of CA in the IUL of children with SUCP. We studied the extent of flexor/extensor CA induced at different velocities of active elbow extension in the IUL of children with SUCP and in the upper limbs of TD children. We developed an approach based on methods that split the movement into the phases of acceleration and deceleration and hypothesized that CA would differ depending on the movement phase. We also expected to find increases in CA with increasing movement speed during active elbow extensions of the IUL.

The second aim was to compare the patterns of activation of the biceps and the brachioradialis muscles during active elbow extension movements.

## 2. Methods

### 2.1. Experimentation

#### 2.1.1. Participants

Thirteen children with SUCP (seven males, mean age = 9.1 years, Standard Deviation (SD) = 2.0, range 6.2–12.5) took part in the experiment. Exclusion criteria for the SUCP participants were: botulinum toxin injections within the previous six months or previous surgery of the upper limbs, inability to fully understand or perform the tasks. Table 1 lists the demographic and clinical data for the children with SUCP (Bohannon and Smith, 1987; Eliasson et al., 2006).

Thirteen TD children (eight males, mean age = 9.5 years, SD = 1.9, range 6.3–12.9) were recruited as a control group. The exclusion criterion for the TD children was previous surgery of the upper limbs. The Research Ethics Board of Sainte-Justine Hospital approved the study and the children’s parents or guardians gave informed consent.

#### 2.1.2. Experimental set-up

**Kinematics:** Upper limb kinematics were assessed using an optoelectronic motion analysis system equipped with twelve infra-

**Table 1**

Demographic and clinical data for the children with SUCP. The Modified Ashworth Scale (MAS) was used to evaluate spasticity [0: none, 4: severe] (Bohannon and Smith, 1987) and the Manual Ability Classification System (MACS) was used to evaluate upper limb function (1: quite good, 5: very impaired) (Eliasson et al., 2006). **Abbreviations:** F-female. IUL-involved upper limb. M-Male. MACS-Manual Ability Classification System. MAS-Modified Ashworth Scale. SUCP-Spastic Unilateral Cerebral Palsy.

Children with SUCP	Age (years)	Gender	MAS for the IUL Flexors	MACS score
1	8.3	F	2	2
2	6.8	M	0	1
3	7.5	M	1+	2
4	9.1	F	1	1
5	9.3	M	1+	2
6	11.3	M	1	1
7	8.9	M	1+	3
8	6.2	F	0	1
9	8.2	M	1+	3
10	10.8	F	1	2
11	12	F	0	2
12	7.8	F	1	2
13	12.5	M	1	2

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