Connecting the dots: Musculoskeletal adaptation in cerebral palsy

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Flexor carpi ulnaris tenotomy alone does not eliminate its contribution to wrist torque.

Why is joint range of motion limited in cerebral palsy patients?

Intramuscular connective tissue differences between spastic cerebral palsy and healthy muscle: a mechanical and histological study.

General Introduction

Spasticity in affected limbs results in substantial torsional adaptations in ulna and radius of patients with cerebral palsy.

Biceps brachii can add to performance of tasks requiring supination in cerebral palsy patients.

General Discussion

Long-term results of lateral band translocation for the correction of swan neck deformity in cerebral palsy.

List of Abbreviations

References

English Summary

Nederlandse Samenvatting
Connecting the dots: musculoskeletal adaptation in cerebral palsy

Patients with hemiplegic cerebral palsy (CP) cope with functional impairment due to movement limitations in one of their arms as a result of pathological motor control. Their inability to extend the wrist and elbow and rotate the forearm hinders them to do things we perform effortlessly. In this thesis I will discuss the results of 5 years of research investigating the musculoskeletal adaptations from which movement limitations in CP may originate. In Chapter 1, the rationale behind the studies in this thesis is explained. In this chapter, an adjusted version of the simple feedback loop in which musculoskeletal structures and movement performance interact with each other is proposed.

In Chapter 2, we try to get an understanding why the movement limitations develop in cerebral palsy. Knowledge of the mechanisms that affect the development of these so-called “contractures” will affect the selection of patients suitable for surgical treatment as well as the choice for specific surgical procedures. The generally accepted hypothesis in patients with spastic cerebral palsy is that the hyper-excitability of the stretch reflex combined with an increased muscle tone result in extreme angles of the involved joints at rest. Ultimately, these extreme joint angles are thought to result in fixed joint postures. There is no consensus in the literature concerning the pathophysiology of this process. Several hypotheses associated with inactivity and overactivity have been tested by examining the secondary changes in spastic muscle and its surrounding tissue. All hypotheses implicate different secondary changes that consequently require different clinical approaches. In this chapter, the different hypotheses concerning the development of limited joint range of motion in cerebral palsy are discussed in relation to their secondary changes on the musculoskeletal system.

In Chapter 3, using biopsies, we compared mechanical as well as histological properties of flexor carpi ulnaris muscle (FCU) from CP patients (n=29) and healthy controls (n=10). The sarcomere slack length (mean 2.5µm, SEM 0.05) and slope of the normalized sarcomere length-tension characteristics of spastic fascicle segments and single myofibre segments were not different from those of control muscle. Fibre
type distribution also showed no significant differences. Fibre size was significantly smaller (1933 \( \mu m^2 \), SEM 190) in spastic muscle than in controls (2572 \( \mu m^2 \), SEM 322). However, correlation analysis indicates that a major part (57.8%) of this difference is explained by age, rather than by the affliction. Quantities of endomysial and perimysial networks were unchanged with one exception: a three-fold thickening of the tertiary perimysium, i.e. the connective tissue reinforcement of neurovascular tissues penetrating the muscle. These results are taken as indications of enhanced myofascial loads on FCU that may contribute to the etiology of limitation of movement at the wrist in CP and the characteristic wrist position of patients.

**Chapter 4** describes a study in which is determined whether connective tissue surrounding the FCU is strong and stiff enough to transmit forces from the muscle to the wrist joint. FCU muscle tenotomy and transfer to the extensor side of the wrist are common procedures used to improve wrist position and dexterity in patients with cerebral palsy. Intra-operatively, we determined in vivo maximal wrist torque in hemiplegic cerebral palsy patients (n=15, mean age 17 years) in three conditions: 1) with the arm and the muscle intact; 2) after tenotomy of the flexor carpi ulnaris just proximal to the pisiform bone, with complete release from its insertion; and 3) after careful dissection of the belly of the muscle from its fascial surroundings up until approximately halfway its length. After tenotomy of the flexor carpi ulnaris muscle, the maximal wrist torque decreased with 18% whereas dissection of the muscle resulted in an additional decrease of 16%. In this chapter, we conclude that despite of the tenotomy of its distal tendon, the flexor carpi ulnaris still contributes to the flexion torque at the wrist through myofascial force transmission. Quantification of this phenomenon will help in the study of the effects of fascial dissection on the functional results of tendon transfer surgery.

The objective of the study described in **Chapter 5** was to evaluate the influence of longstanding wrist flexion, ulnar deviation, and forearm pronation due to spasticity on the bone geometries of radius and ulna. Furthermore, the hypothetical influence of these deformities on potential maximal moment balance for forearm rotation was modeled. Bone volume, length and geometrical measures were determined in
hemiplegic cerebral palsy patients (n=5) and controls (n=5). Bilateral differences between the spastic arm and the healthy side were compared to bilateral differences between dominant and non-dominant side in the healthy controls. Patients showed significantly smaller (radius: 41.6%; ulna: 32.9%) and shorter (radius: 9.1%; ulna: 8.4%) forearm bones in the non-dominant arm than the dominant arm compared to controls (radius: 2.4%; ulna 2.5% and radius: 1.5%; ulna: 1.0% respectively). Furthermore, patients showed a significantly higher mean torsion angle difference (radius: 24.1°; ulna: 26.2°) in both forearm bones between arms than controls (radius: 2.0°; ulna: 1.0°). The decreased loading and unbalanced loading of the bones in the spastic forearm causes these bones to be substantially smaller and have a torsion that is approximately 25° larger compared to the contralateral healthy arm. Torsion in the bones of the spastic forearm is likely to influence potential maximal moment balance and with that forearm rotation function. In this chapter it is suggested that torsion of the forearm bones in the spastic arm should be considered when evaluating movement limitations, planning treatment and evaluating outcome of treatment in the upper extremity of CP patients.

In Chapter 6 it was assessed whether cerebral palsy patients can use biceps brachii for supination during movement tasks requiring supination and pronation. 3D upper extremity kinematic and EMG-data of twelve patients (mean age 13 y 8 mo ± 36 mo) were compared to 10 healthy age-matched controls. Significant difference in biceps brachii activation between maximal isolated pronation and supination in both groups showed that it is possible for CP patients to use biceps brachii for supination. Performance of reach-to-grasp with either pronation or supination showed similar activation patterns as during isolated tasks in both groups, although increased biceps brachii activation likely also hampered performance of reach-to-grasp in the patient group by causing increased, and possibly unwanted elbow flexion. However, the functional effect of this flexion for supination purposes cannot be ruled out. Therefore, it is concluded that one should be cautious with simply weakening biceps brachii when the purpose is to improve functional reach. Ideally treatment might
focus more on changing the flexion moment/supination moment ratio of biceps toward a stronger supination function.

Finally, in Chapter 7 we tried to connect the dots between the different studies in this thesis by discussing the results of the different studies with respect to the overall aim of this thesis. The outcomes of the different studies described in this thesis emphasize that this is a multi-dimensional problem. The challenges in improving treatment lie in finding the starting point for the changes in tissue structure and mechanics and unraveling the interactions between these characteristics of all tissues. The multidisciplinary approach that is already used in treatment of movement limitations in cerebral palsy should therefore be extended in fundamental research. However, the key to successful treatment of movement limitations in this patient group might be longitudinal studies that clarify both healthy musculoskeletal development and the way in which this development is affected by the altered motor control. Knowledge on the development of musculoskeletal structures could give us direction where to aim interventions that might reverse and prevent changes that lead to movement limitations in these patients.

The aim of the study described in the Appendix was to evaluate the long-term effect of lateral band translocation for correcting swan neck deformity in patients with cerebral palsy at a minimum follow-up of 5 years. Swan neck deformities of 62 fingers were corrected using a modified lateral band translocation. At 1-year and 5-year follow-up, any recurrence of hyperextension was recorded through unconstrained evaluation. Active extension of the proximal interphalangeal joint beyond 0 degree was considered a recurrence. Correction was successful for 84% of the operated fingers at 1-year follow-up. After 5 years, the success rate had decreased to 60%. Furthermore, no relationship was found between any of the concomitant surgical procedures and the number of patients with recurrences. The long-term result of lateral band translocation is disappointing in our series, and it should not be advocated as a procedure with a long-lasting success in patients with cerebral palsy.