SUMMARY

Children with spastic cerebral palsy (CP) often develop contractures in joints due to muscle shortening or increased muscle stiffness. This may lead to limitations in joint range of motion (ROM). The most common contracture developing in children with CP is the reduction of ankle-foot dorsiflexion (A-Fdf) ROM (measured with extended knees) due to a shortened or stiffer m. gastrocnemius (GAS). The reduced dorsiflexion ROM results in an impaired gait. In particular children walking with excessive ankle-foot plantar flexion and flexed knees show decreased A-Fdf ROM. A knee-ankle-foot orthosis (KAFO) worn at rest, which includes ankle and knee fixation to apply strain on the GAS, is commonly prescribed in clinical care. It is expected that GAS length can be maintained or even increased by the strain because a muscle may be able to adapt its optimum length to the joint position in which the muscle is frequently active. However, very little is known regarding the efficacy of wearing a KAFO at rest and whether the assumptions regarding the underlying working mechanisms are correct. The primary aim of this thesis is to quantify potential effects of treatment with knee-ankle-foot orthoses over time in children with spastic CP. In addition, we aimed 1) to investigate whether parent reported KAFO wearing time can be considered as a valid indicator and 2) to assess effects of affected A-Fdf ROM on gait kinematics in children with CP walking with plantar flexion in the ankle and flexed knees. The aims are discussed in chapter 1.

Chapter 2 describes the protocol of a single blinded randomized controlled trial investigating the efficacy of wearing KAFO’s in children with spastic CP to prevent a decrease in A-Fdf ROM. It was aimed to follow three groups of children with spastic CP for one year. One group was treated with a static KAFO (fixed knee at 180° extension and fixed ankle at 0° dorsiflexion) and usual care and one group was treated with a dynamic KAFO (fixed knee at 180° extension and a dynamic ankle applied with an Ultraflex® power unit) and usual care. The third group was included as a control group and received usual care only (physical therapy, manual stretching). KAFO’s had to be worn for at least 6h every other night. For all children participating, KAFO treatment to prevent a decrease in A-Fdf ROM was indicated because they were at risk for reduction of A-Fdf ROM due to their medical history. At baseline and after 3, 6, 9 and 12 months, A-Fdf ROM was measured using a custom designed hand-held dynamometer. In addition, measurements to obtain information about gait kinematics, gross motor function, wearing time and complaints were performed. In a subgroup of patients, information about morphological parameters was collected using a 3D imaging technique. Results of morphological measurements are not presented in this thesis.
Chapter 3 describes results of the randomized controlled trial. 28 children (15 in the dynamic KAFO group and 13 in control group) with spastic CP and an age between 4 and 16 years old participated in the study. The static KAFO group had to be removed from the study design after the inclusion of some participants as they were not able to wear the KAFO because they experienced too much pain during wearing. Regarding the comparison between the control group and dynamic KAFO group, no effect was found in the decrease of A-Fdf ROM, gait kinematics and gross motor function. Other interesting outcomes were that: 1) the expected reduction of A-Fdf ROM over time in the control group was only statistically significant at 6 months, but not at 9 or 12 months, 2) 11 participants (4 in the experimental group and 7 in the control group) did not complete all five measurements, as they needed additional treatment, 3) wearing time of the dynamic KAFO was low and 4) all participants of the experimental group complained about pain and/or sleeping problems while wearing the KAFO. It was concluded that a dynamic KAFO was poorly tolerated and not beneficial in preventing a reduction in A-Fdf ROM, at least with limited use.

Chapter 4 presents the results of a study comparing the measurements of parent-reported wearing time (collected by questionnaires) and those of objectively measured wearing time (collected by temperature sensor-data-loggers attached to the KAFO’s). Although mean difference between the two measured wearing times was low, there was a high inter-individual variation between parent-reported and objectively measured wearing time. Therefore, objective measurement methods to measure KAFO wearing time are recommended. Differences in parent-reported wearing time may bias results of efficacy studies and hinder the possibility to investigate the relation between wearing time and treatment efficacy.

The study described in chapter 5 investigated effects of changes in A-Fdf ROM on gait kinematics in 10 children with spastic CP, walking with knee flexion and ankle-foot plantar flexion in mid stance. Although an effect of KAFO treatment could not be shown, participants of the randomized controlled trial showed large individual variation in A-Fdf ROM over time. This study showed that when A-Fdf ROM changed over time, this resulted in changed knee extension in mid stance of gait, rather than a change in A-Fdf in mid stance of gait. This finding is likely due to the fact that the GAS (which length change causes decreased A-Fdf ROM) is a bi-articular muscle generating a moment over both ankle and knee. This study shows that effects of involved muscles have to play a role when selecting treatment options improving gait pattern of children with spastic CP. The effect of the GAS needs to be taken into account when treating patients with excessive ankle-foot plantar flexion and knee flexion.
Chapter 6 includes a discussion of the findings of the research as a whole. Based on the results of the different studies it is concluded that the lack of effect of the dynamic KAFO may be caused by the low KAFO wearing time due to low tolerance and/or by the inability of the KAFO to apply a sufficient high strain on the GAS. It is therefore suggested that a KAFO with modified design will be more effective. A KAFO allowing 20° knee flexion is presumed to be better tolerated. However, to be able to assess whether such a KAFO strains the GAS sufficiently, requires further research. It should be evaluated whether the KAFO stabilizes the foot bones of hind- and midfoot sufficiently to allow application strain on the GAS, but also whether there are local strain amplifications which may be necessary for lengthening of the muscle fibers (i.e. to increase the number of sarcomeres in series). In addition, it is presumed that strength training during KAFO wearing is required to stimulate addition of sarcomeres in series within muscle fibers and to prevent atrophy (i.e. reduction in the physiological cross-sectional area). In children with spastic CP, the rate of addition of sarcomeres in series may be reduced compared to that in typical developing children. Moreover, physiological cross-sectional area may be reduced due to immobilization of the GAS due to fixed knee flexion while wearing a KAFO. Strength training may prevent this atrophy as well. The efficacy of treatment with a modified treatment needs to be tested while measuring wearing time using with objective measurement techniques. If the suggested modified KAFO is better tolerated and more effective than the KAFO tested in the present study, it may be a promising approach to improve A-Fdf ROM and to reduce knee flexion in gait in children walking with excessive knee flexion and ankle-foot plantar flexion.