Summary

Cerebral palsy (CP) is a group of disorders of the development of movement and posture, causing activity limitations, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. Problems with walking are common, and levels of physical activity and fitness are lower compared to typically developing children. The benefits of physical activity on fitness and health, widely recognized for the general population, are suggested to be especially important for children with a physical disability, such as children with CP. In this thesis the factors that might influence the physical activity level of children with CP were examined. Thereby, the physical activity level of children with CP was compared to typically developing children (TDC) using step activity monitors and heart rate monitors. Furthermore, the effectiveness of a six-month physical activity stimulation program was examined, which included counseling through motivational interviewing, home-based physiotherapy, and fitness training for children with CP.

Physical activity

Chapter 2 reports on the results of a cross-sectional study on the walking activity of sixty-two children with spastic CP in the age of 7-13 years, who walk without aids (classified as Gross Motor Function Classification System [GMFCS] level I and II) or use walking aids (GMFCS III). The aim of the study was to describe walking activity, and to identify the association of disease, personal, and environmental characteristics with walking activity, while body height was taken into account. Walking activity was assessed during school and weekend days, using an accelerometer (StepWatch™) that registers strides per minute. Outcomes were the daily number of strides, and the time spent at different stride rate levels. Disease characteristics were GMFCS level and limb distribution, personal characteristics comprised age, gender, attitude towards sports and athletic competence, and environmental characteristics comprised school type, siblings, parental educational level and sports club participation. On school days, reduced walking activity was associated with a higher GMFCS level, bilateral spastic CP, and a higher age. The explained variance (R^2) ranged from 43 to 53 % for the different outcomes of walking activity. On weekend days, reduced walking activity was associated with bilateral spastic CP, a higher age, and lack of sports club participation

(R^2 ranged from 21 to 42 %). No associations were found with the other personal and environmental characteristics. In addition, walking activity was considerably lower on weekend days than on school days ($p<0.001$). These findings demonstrate that older children with bilateral spastic CP and a higher GMFCS level are at risk for inactivity, and should be targeted for intervention. In particular, physical activity at the weekend should be promoted.

In **Chapter 3** the walking activity between Dutch and American children with CP, and age- and gender matched TDC, was cross-sectionally compared. Children with CP were classified as GMFCS level I-III and included in the study when they were 7-12 years old. Walking activity was registered with the StepWatch™. Outcomes were the daily number of strides, and the time spent inactive and at low, moderate and high stride rate levels. On all outcomes walking activity of Dutch and American TDC was comparable between countries. However, it was different for children with CP. Dutch children in GMFCS I-II (walking without aids) showed less walking activity, particularly at high stride rates, compared to their American counterparts. In contrast, Dutch children in GMFCS III (using walking aids) showed more walking activity, especially at moderate stride rates, than their American counterparts. In both countries the walking activity of children with CP was lower than TDC. Differences between countries vary between TDC and children with CP walking with or without aids. This may indicate that the environment affects children with CP and TDC in a different way.

In **Chapter 4** the walking activity and heart rate responses of 7- to 14-year old children with CP (GMFCS I-III), and age- and gender matched TDC, were cross-sectionally compared. Children simultaneously wore a StepWatch™ and a heart rate monitor for a period of three days. Daily stride rate activity and heart rate reserve were each divided in five intensity categories (inactive, low, moderate, high, very high). The time spent in each stride rate and heart rate reserve category were compared between children with CP and TDC. Thereby, the mean heart rate reserve at each stride rate category was compared between groups. Children with CP spent more time inactive, and less time at all daily stride activity categories than TDC, but the active time spent at each heart rate reserve category was similar between groups. Mean heart rate reserve of

TDC and children in GMFMCS I-II was similar for each stride rate category. For children in GMFCS III mean heart rate reserve was higher than in TDC at all stride rate categories, except for the highest stride rate category. These results demonstrate that stride rate activity levels reflect a similar effort of walking of TDC and children in GMFCS I-II (walking without aids), and that children in GMFCS III (using walking aids) have a higher effort of walking than TDC. These results also show that the StepWatch™ and a heart rate monitor measure different aspects of daily physical activity. Stride rate activity measured with a StepWatch™ might preferably be used for monitoring daily walking activity, while HRR intensity derived from heart rate monitoring may better suited for capturing vigorous exercise.

Physical activity stimulation program

The study protocol in **Chapter 5** extensively describes the rationale, design, intervention, and outcome measures of the six-month physical activity stimulation program called "LEARN 2 MOVE 7-12". It was hypothesized that the physical activity stimulation program would have added value for improving physical activity compared to regular pediatric physiotherapy. Children with spastic CP in the age of 7 to 13 years, who were able to walk independently (GMFCS I-III) were invited to participate. Children were recruited via local physical therapy practices and special schools for children with a disability. They were randomly allocated to the intervention (physical activity stimulation program) or the control group (regular pediatric physiotherapy). The physical activity stimulation program consisted of a six-month lifestyle intervention involving counseling and home-based physiotherapy, parallel to a four-month group fitness training. Counseling of the family took place at the child's home and through follow-up telephone calls, using the motivational interviewing technique. Home-based physiotherapy was individually tailored to the child's needs and aimed at practising mobility-related activities in the daily situation. The one-hour fitness training sessions (first two months twice a week, and third and fourth months once a week) involved lower-extremity muscle strengthening exercises, as well as anaerobic exercises. The training load was progressively increased over the four-month training period. Data was collected at baseline, after four months fitness training, at the end of the intervention (at 6 months), and at six months follow-up by an assessor blinded for group allocation. The primary outcome measure physical

activity included walking activity measured with an accelerometer (StepWatch™) and parent-reported physical activity. Secondary outcome measures included mobility capacity (gross motor capacity [GMFM-66]), walking capacity, and functional muscle strength), fitness, fatigue, attitude towards sports, social participation in domestic life and in recreation and leisure, self-perception and quality of life. To determine the intervention effect, intention-to-treat analyses were performed using linear regression analyses with baseline correction ($p<0.05$). The results of the study are described in chapter 6 and 7.

Chapter 6 describes the effectiveness of the physical activity stimulation program on physical activity, mobility capacity, fitness, fatigue and attitude towards sports. Fifty children with spastic CP were included, of whom forty-six completed the study (26 male, mean age 9y 8mo, SD 1y 8mo). After correcting for multiple testing, no statistically significant intervention effects were found for physical activity, or the other outcome measures, neither at the end of the fitness training program, the end of the intervention, nor at follow-up. Borderline significant results were found for one of the physical activity outcomes (parent-reported time at moderate-vigorous intensity: $p=0.04$) and gross motor capacity (GMFM-66: $p=0.03$) at the end of the intervention, but not at follow-up. These results demonstrate that the combination of counseling through motivational interviewing, home-based physiotherapy and fitness training was not effective for improving physical activity of children with CP. Consequently, the hypothesis that these different treatment components would work synergistically for improving physical activity could not be confirmed. Further research should examine how each separate component of the intervention may contribute to improving physical activity in this population. Possible explanations for the lack of an intervention effect may be insufficient contrast between groups (all families were interested in expanding physical activity), or the relatively short duration of the program (six months) to elicit changes in a complex behaviour like physical activity. Whether a longer counselling period with periodical attention to physical activity may be needed to improve physical activity in children with CP should be examined in further research.

In **Chapter 7** the effectiveness of the physical activity stimulation program on social participation in domestic life and recreation and leisure, and on self-perception and quality of life was reported. After correcting for multiple testing, there were no statistically significant effects for social participation in recreation and leisure, and self-perception at the end of the intervention and at, or for quality of life at follow-up. A borderline significant result was found for social participation in domestic life at follow-up ($p=0.03$) reflected by an improved accomplishment of activities in and around the home, but not at the end of the intervention. These results show that the physical activity stimulation program was not effective in improving social participation, self-perception and quality of life when compared to regular pediatric physiotherapy, but that the program did show potential in improving social participation in domestic life over the longer term. This suggests that barriers to expand the activity level in the home may take time and effort to overcome, but also that expanding activities in and around the home may be a more practical and feasible alternative for children with CP than expanding activities in the community such as sports. This may provide directions for the development of future physical activity stimulation programs.