General discussion
Understanding functional prognosis in children and adolescents with cerebral palsy (CP) is indispensable to improving pediatric rehabilitation treatment. An additional important consideration is that parents and children with CP wish to know what to expect from daily life as they grow older. The aims of this thesis were to describe the course of (daily) activities and to determine relationships with disease characteristics, personal factors and environmental factors in children and adolescents with CP. The second aim was to investigate their physical activity level and get insight in the experienced activity and participation problems of the children and adolescents with CP; and to examine whether the utilization of healthcare services matched the problems experienced.

Achieving these goals was facilitated by the participation of 110 children and their parents in a three year follow-up study with annual measurements. This chapter commences with a recapitulation and discussion of the main findings and clinical implications of our study, followed by proposals for future research.

Development of mobility

By clinical observation a part of children with CP seem to deteriorate around the age of 10 to 14 years of age. The growth spurt and increase in body weight that take place during puberty in children with CP is thought to lead to a deterioration in mobility.\textsuperscript{1-3} This was the impetus for the PERRIN 9–16 years study. However, our study could not confirm a deterioration in mobility as a result of skeletal maturation (see Chapter 2).\textsuperscript{4} Hanna et al. showed, in a longitudinal study, a decline in mobility for GMFCS III to V at approximately 7 years of age.\textsuperscript{5} A more recent longitudinal analyses of a Dutch population of individuals with CP (including our study population) showed the existence of five distinct patterns for gross motor development by level of severity of CP.\textsuperscript{6} Although that study indicated that mobility does not deteriorate but remains stable in adolescence, some individuals showed deterioration in mobility. The group of young adults (aged 16 to 24 years) in that study lacked individuals with an intellectual disability (IQ<70) as this had been an exclusion criterion in the original study.\textsuperscript{7} Therefore, young adults at GMFCS levels III, IV, or V are underrepresented and their mobility parameters probably overestimated. As stated above, our study showed that the deterioration that was found in some children was not caused by a growth spurt, but could be influenced by factors other than skeletal maturation (e.g., severity of CP or intellectual disability). As described in Chapter 2, we found a high prevalence of children whose skeletal age was either advanced or delayed in relation to their chronological age, and we showed that skeletal age in girls with CP was on average significantly higher than their
chronological age. Recently, a study on the long-term outcome of single-event multilevel surgery in spastic diplegic CP showed that girls maintained an enhanced level of walking, while walking ability in boys deteriorated. This finding suggests that sex affects treatment response in walking ability, and that skeletal maturity, which is known to be advanced in girls, might explain the difference in treatment response between girls and boys.

In Chapter 3, we described the course of mobility performance (measured with the VABS) in comparison with the course of mobility capacity (measured with the GMFM) for different GMFCS levels. While we expected that capacity would exceed performance, its relationship to the severity of CP was unknown. Our results showed a less favorable course of mobility performance for children at GMFCS levels II, IV, and V, compared with children with mild CP (GMFCS level I) over a period of 3 years. Although the measurement instruments of performance and capacity could not be compared directly, the results suggested that mobility performance of children in GMFCS level III was higher in relation to their capacity than mobility performance of children in other GMFCS levels. This could be explained by the fact that these children used walking aids which increased their mobility performance to a higher level than could be expected on the basis of their mobility capacity. The results also suggested that, in children with GMFCS levels IV and V, mobility performance was relatively lower compared to mobility capacity than in children with other GMFCS levels. We were able to demonstrate a positive longitudinal relationship between mobility capacity and mobility performance. This positive longitudinal relationship plausibly suggests that training mobility capacity will increase motor performance. Recently, Smits et al. used the term capability in addition to the terms capacity and performance. Capability refers to what an individual can do in a daily environment. They found moderate correlations of change scores for motor capacity and motor capability, motor capacity and motor performance, and motor capability and motor performance in children and adolescents aged 1 to 16 years (including our study population). Vos et al. studied the developmental trajectories of mobility performance and daily activities in children and young adults with CP (including our study population), and explored the influence of gross motor function and intellectual disability on these trajectories. Results showed that the developmental trajectories of mobility performance differed by GMFCS level, whereas intellectual disability had no influence. These results differ from findings on the developmental trajectories of mobility capacity reported in the same population (GMFCS I–V aged 1–22 or 24 years) in which inclining development peaked much earlier, at age 7 to 9 years (depending on the GMFCS level). The continuing incline of mobility performance suggests that when mobility capacity stabilizes after childhood, it is important to continue rehabilitation that focuses on valuable and meaningful activities in the context of the person's daily life.
Clinical implications for development of mobility

When health care professionals carry out interventions that depend on growth status (i.e. orthopedic or multilevel surgery), it might be advised to determine skeletal age, as a large percentage of girls with CP will reach the end of growth at an earlier age than might be expected from their chronological age. Although some adolescents might show deterioration in mobility during adolescence, in information to parents and adolescents with CP it is important to indicate that mobility may deteriorate, but is not expected to deteriorate as a result of skeletal maturation.

The findings of the longitudinal study on mobility performance suggested that training mobility capacity in the context of person's daily life is important for increasing motor performance. Furthermore, the course of mobility performance in relation to mobility capacity in children in GMFCS levels IV and V seems to be less favorable. For those adolescents, who depend to a larger extent on aids, greater attention should be paid to interventions that focus on the child's environment and mobility equipment. In relation to improving the mobility capability and performance of adolescents with CP, Smits et al. suggested that they would benefit from a focus on environmental factors (e.g., physical environment, but also social support) and personal factors (e.g., the child's motivation). However, as only small changes in mobility capacity and performance were observed for children classified at GMFCS levels IV and V, the clinical focus for these subgroups may emphasize other aspects (e.g., quality of life, participation in daily life).

Recommendations for future research on development of mobility

Our findings on the development of mobility in adolescents with CP lead to certain recommendations for future research. As the follow-up of our study was relatively short, it is recommended that participants are again measured within a few years to follow the development of mobility in young adults. In-depth analyses of sex-specific factors representing development in puberty – such as increase in height, changes in body mass index, Tanner stage, and hormone levels – might explain changes in mobility capacity in adolescence. The growing evidence of differences in growth and treatment response between girls and boys with CP suggests that the influence of skeletal maturity and perhaps some other gender-specific differences in children should not be underestimated. Therefore, further research on the possible influence of these factors in adolescence might provide insights that are relevant to individual adolescents at risk for deterioration of mobility in puberty. In order to better understand the longitudinal relationship between mobility capacity and mobility performance, it is important to use an instrument that measures mobility.
capacity and performance similarly, such as the Activities Scale for Kids. More research is also needed on the role of the environment (physical and social support) and the use of mobility equipment on mobility performance over time.

**Manual ability and daily activities**

The results of our cross-sectional study in Chapter 4 confirmed that a large percentage of adolescents with CP show limited manual ability and that these limitations were strongly related to limitations in daily activities. These findings were confirmed by a recent study on the influence of hand skills (measured with the Assessment of Children's Hand Skills) on self-care (measured with VABS) in younger typically developing children and in children with disabilities (including CP). As manual ability, classified with the MACS, was limited (MACS II–V) in 59% of the adolescents participating in the present thesis, and was strongly associated with daily activities, therapy programs are expected to improve manual ability in these children. A recently published systematic review of systematic reviews of interventions for children with CP showed several “green light” interventions to improve activities, including home programs and goal-directed or functional training. “Green light” interventions were defined as those which were supported by high or moderate quality evidence and high recommendation according to GRADE and Evidence Alert Traffic Light System. An example of a “green light” intervention to improve manual ability in younger children with CP is ‘Constrained Induced Movement Therapy’ (CIMT) and bimanual training. Both interventions can be used to improve manual performance. However, the learning curve of manual performance during treatment with CIMT and bimanual training was more favorable in children younger than 5 years of age. Although not yet supported by meaningful evidence, new treatments such as robot-assisted and computer-enhanced therapies might be complementary to established treatments. In Chapter 4 we used the MACS as outcome measure. The MACS was developed after the start of the PERRIN study, and was therefore only included in the last measurement and not measured over time. Since then, a large number of studies of manual ability in children with CP have been performed, strengthening the evidence of MACS as being a valid and useful classification of children's manual ability. More recently, one study suggested that MACS levels are stable over time and that the classification has predictive value.

**Clinical implications on manual ability and daily activities**

Based on the findings above, it is recommended that measurements of manual ability should be included in the regular assessment of adolescents with CP, and that these measures of
manual ability should be incorporated into rehabilitation treatment for evaluation. In daily practice the MACS classification could be used to give an indication of limitations in self-care, and the ABILHAND-Kids score could be an indicator of limitations in domestic life.\textsuperscript{15} It is recommended to use CIMT or bimanual training in order to improve manual ability,\textsuperscript{17} although optimal dosage of training in children older than 5 years of age is not yet known.\textsuperscript{20}

**Recommendations for future research on manual ability and daily activities**

To understand whether improvements in manual ability, for example following treatment with CIMT or bimanual training, will also lead to improvements in the performance of daily activities over time, a longitudinal study or a randomized clinical trial with a long-term follow-up is recommended.

**Physical activities and experienced problems and healthcare utilization**

In Chapter 5, the results of our study showed that a large proportion (78%) of adolescents with CP participated weekly in sports activities. However, according to the Dutch norm for physical activity, 89% of the adolescents with CP were insufficiently physically active in relation to sports and cycling. Higher age, female gender, and hip dysplasia were significantly associated with a lower level of physical activity. Physical activity contributes to development of physical fitness and is therefore considered to be important for children with CP. Maintaining physical activity at an adequate level is especially important in children with mobility problems, because it improves health, reduces secondary conditions, and enhances quality of life.\textsuperscript{27} Stimulating physical activity is important, however a physical activity stimulation program in The Netherlands for children with CP (aged 7–12 years) showed no statistically significant improvements in physical activity.\textsuperscript{28,29} Likewise, a lifestyle intervention program in adolescents and young adults with CP showed that fatigue, bodily pain, mental health and social support can be improved,\textsuperscript{30} but the program was ineffective in eliciting a behavioral change to more favorable physical activity.\textsuperscript{31} Consideration of these results and other study outcomes resulted in the idea that intervention might be more effective when focusing on decreasing unhealthy behaviors rather than increasing healthy behaviors.\textsuperscript{32} Thus, interventions focusing on decreasing sedentary behavior may be more effective than current interventions emphasizing increase of physical activity.\textsuperscript{33,34} Another possibility might be to change the environment in order to challenge physical activity.\textsuperscript{35} Positive associations between urban planning and physical activity have been demonstrated in typically developing children.\textsuperscript{36} In addition, it is known that social environmental influences, such as being allowed to play outside, parental activity rules, and family encouragement, are related to enhanced
physical activity in typically developing children. Environmental and social barriers which were observed during the counseling module in a physical activity stimulation program in children with CP can serve as a possible starting point for interventions.

The study described in Chapter 6 showed that 79% of adolescents with CP experienced one or more activity and participation problems in daily life. Problems were mostly experienced in the mobility domain, but problems were also experienced in education, recreation and leisure, self-care and interpersonal relationships. There seems to be a particular discrepancy between problems experienced in education, play/hobbies, and interpersonal interactions/relationships and the healthcare services provided. As the problems experienced by these adolescents might shift as they mature, the transition to young adulthood should be taken into account and appropriately coordinated by healthcare teams specialized in young adults. The goal of rehabilitation treatment is achieving the optimal level of participation, support for participation problems in adolescence is therefore essential. Several studies have stressed the importance of adequate care for young adults with physical disabilities and have promoted specialized rehabilitation teams for young adults with a physical disability. However, these studies also indicated that there is a need for improvement in the healthcare provided. A recent study on transition and the needs of adolescents with CP highlighted the importance of ensuring that the transition to adult rehabilitation remains flexible and is not determined by a given biological age. Our study suggests that the provision of healthcare services in domains that will become even more important in adulthood (e.g. interpersonal relationships) are already lacking at earlier ages.

Clinical implications for physical activities and experienced problems and healthcare utilization

Professionals, but also parents, should be aware of the low physical activity levels of children with CP, especially in older adolescents, girls, and adolescents with hip dysplasia, and should promote physical activity – preferably in everyday settings whenever possible. For children with hip dysplasia, special attention should be paid to finding alternative sports activities (e.g. hand cycling) that put less strain on the hips but do promote improved physical fitness. Recently, Lauruschkus et al. also found reduced physical activity in the more severely affected children and emphasized the need of specific attention for these children when planning interventions for increasing physical activity. Furthermore, therapists and physicians should be aware of a possible mismatch between experienced problems and healthcare provision, and thus place the appropriate emphasis on providing the necessary services. In addition, these healthcare professionals should anticipate the new activity and participation problems that may arise as adolescents progress into young adulthood.
Recommendations for future research on physical activities and experienced problems and healthcare utilization

A positive longitudinal relationship between mobility capacity and mobility performance has been demonstrated in this thesis. The environment and social support is suggested to play an important role in this relationship, but also in relation to enhanced physical activity. Therefore, further research on environmental and social influences on physical activity of adolescents with CP might provide insight into whether and how more active behavior could be stimulated or sedentary behavior could be discouraged.

In our study of activity and participation problems and healthcare utilization we did not ask whether there was a specific need for healthcare services, but compared the adolescent-experienced problems with the reported healthcare utilization. Therefore, we were not able to investigate if there were unmet needs in the healthcare services offered. Future research on these specific needs in a larger group of adolescents with CP is recommended to provide better insight into a further mismatch between needs and healthcare provision.
REFERENCES


