Summary
During the period of growth and development from childhood to adulthood, adolescents are mainly concerned with consolidating identity, achieving independence from parents, establishing relationships, and finding proper education. Chronic disability during this important life phase inevitably has implications for a child’s development, and in the case of adolescents with cerebral palsy (CP), a variety of secondary musculoskeletal problems such as muscle shortening or joint contractures may develop. These musculoskeletal problems and the physical changes that occur during adolescence may affect the course of motor functioning and may influence their daily activities and social participation.

This thesis describes the findings of the PERRIN CP 9–16 year study (longitudinal study of children and adolescents with CP aged 9 to 16 years). The aims of this study were to describe the course of daily activities and to determine its relationship with disease characteristics, personal factors and environmental factors in children and adolescents 9 to 16 years. 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.

Chapter 1 comprises the introduction, the aims and the design of the study presented in this thesis.

Chapters 2, 3 and 4 relate to the primary aim of this thesis, i.e. the relationship between disease characteristics, personal factors, environmental factors and the course of daily activities in children and adolescents with CP aged 9 to 16 years.

In Chapter 2, we describe skeletal maturation in relation to chronological age in 100 children with CP aged 9 to 13 years (37 females, 63 males; age 9, 11, or 13 years; 73 ambulant, 27 non-ambulant) over a period of 3 years, and its relationship with mobility. Skeletal age was determined based on X-rays of the hand (Greulich and Pyle technique) and mobility was measured with the Gross Motor Function Measure-66. A large percentage (13 to 41%) of children with CP had a skeletal age that deviated (advanced as well as delayed) by 1 year or more from their chronological age. While girls with CP showed advanced skeletal age in relation to chronological age during (pre-)puberty, this was not seen in boys. No difference was found in the course of skeletal age in relation to chronological age over a 3-year period between boys or girls or with regard to level of ambulation. Furthermore, we found no evidence that children with CP are at risk of deterioration in mobility as a result of skeletal maturation during puberty. In Chapter 3 we analyze the course of mobility performance and its relationship with mobility capacity in 104 children (66 males, 38 females) with CP (aged 9, 11, and 13 years at the start of the study) over a period of three
years. Mobility performance, ‘what a child does’, was determined using the gross motor skills subscale of the Vineland Adaptive Behavior Scales (VABS), and mobility capacity, ‘what a child can do’, was determined using the Gross Motor Function Measure-66 (GMFM-66). The measurements were performed annually over a period of 3 years. The course of mobility performance in mildly affected children (Gross Motor Function Classification System (GMFCS) level I) was more favorable than in more severely affected children (GMFCS levels II, IV, and V). 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. Therefore, GMFCS level can be used to identify children who may be at risk for deterioration in mobility performance during adolescence. An increase in mobility capacity was significantly related to an improvement in mobility performance over the 3 years. Training mobility capacity in children with CP seems to be important for improving mobility performance. Interventions should also focus on environmental adaptations and improving mobility equipment. A limitation of this study was that the instruments used did not contain the same items on capacity and performance level. Chapter 4 describes the manual ability of adolescents with CP and the relationship of manual ability to daily activities. Manual ability was classified with the Manual Ability Classification System (MACS) and the ABILHAND-Kids. Daily activities were assessed with the VABS subscales for personal and domestic daily living skills. The relationship between manual ability and daily activities was investigated with regression analysis: independent variables were manual ability, disease and personal characteristics. MACS and ABILHAND-Kids were both strongly associated with personal daily activities (explained variance 77% and 84%, respectively) and less strongly with domestic daily activities (explained variance 45% and 62%, respectively). Including other disease characteristics and personal characteristics in the model increased the explained variance of personal daily activities to 91% for both models and the explained variance of domestic daily activities to 68% and 73% for the MACS and ABILHAND-Kids models, respectively. The results of our study confirm that manual ability is limited in a large percentage of adolescents with CP, and that limitations in manual ability are strongly related to limitations in daily activities. The classification or assessment of manual ability should be a regular component of the physical examination in order to guide decisions concerning further treatment.
Chapters 5 and 6 relate to the second aim of this thesis, i.e. the physical activity level and experienced activity and participation problems of adolescents with CP and the match with utilization of healthcare services.

In Chapter 5 we describe the physical activity levels of adolescents with CP and associated factors. Physical activity levels were measured by means of a questionnaire filled in by the parents of 72 adolescents with CP (12–16 years of age) and expressed in METs. The results of this study indicated that a large proportion (78%) of the adolescents with CP participated weekly in sports. However, according to the Dutch norm for physical activity, 89% of the adolescents with CP were insufficiently physically active in sports and cycling. Older age, female gender, and having hip dysplasia were significantly associated with a lower level of physical activity. It is therefore important to promote physical activity, especially among older adolescents with CP (age 14–16 years), girls, and adolescents with hip dysplasia. In Chapter 6 we describe the activity and participation problems experienced by 94 adolescents with CP aged 12 to 16 years and examined the match with utilization of healthcare services. The problems experienced by the children were measured using the Canadian Occupational Performance Measure (COPM) and categorized into activity and participation domains. The utilization of healthcare services was assessed with a questionnaire and matched to the experienced problems. We found 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 in problems experienced in education, play/hobbies and interpersonal interactions/relationships and the utilization of healthcare services in these domains. Therapists and physicians should therefore be aware of a possible mismatch between experienced problems and healthcare provision and give sufficient attention to providing the necessary services.

The general discussion in Chapter 7 starts by recapitulating and discussing the main findings and clinical implications of our study, followed by directions for future research.

Recommendations for future research include a longer follow-up to investigate the development of mobility in young adults. In addition, this thesis also showed that explanatory research on gender differences is desirable. More research is also needed to investigate the role of the environment and the use of mobility equipment on mobility performance over time. To understand whether improvements in manual ability, for example after treatment, will also lead to an improvement in the performance of daily activities over time, a longitudinal study or a randomized clinical trial with a long-term follow-up is recommended. Further
research on environmental influences on physical activity of adolescents with CP might provide insight into methods that encourage more active behavior. Finally, future research on the specific needs in a larger group of adolescents with CP would be recommended to provide better insight into a possible mismatch between experienced problems and healthcare provision.