Summary
This thesis describes the findings of the PERRIN CP 9-16 year study (longitudinal study on functioning and health-related quality of life in children and adolescents with cerebral palsy aged 9 to 16 years) which was part of the PERRIN (Pediatric Rehabilitation Research in the Netherlands) program. The PERRIN CP 9-16 year study is a multicenter, prospective cohort study on 110 children with cerebral palsy and their parents. The children were 9, 11 and 13 at the start of the study and were followed for three years (four yearly measurements). The main aims of the study were:

1. to describe the level and course of activities and participation and to determine its relationship with disease characteristics, personal factors and environmental factors in children and adolescents with CP aged 9 to 16 years; and
2. to investigate their self-esteem and get insight to their health-related quality of life.

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

Chapters 2, 3 and 4 describe the first aim of this thesis, i.e. the level and course of activities and participation on several domains of functioning.

In Chapter 2 the level of activities and participation was investigated at baseline in the domains of mobility, self-care, daily living skills, social functioning and communication, in relation to personal factors and disease characteristics. The results showed that many children with CP had limitations on all domains of functioning. The severity of the CP, measured with the Gross Motor Function Classification System (GMFCS level) was the most important factor among the ones studied that was significantly associated with mobility, self-care and daily living skills. Cognitive impairment and epilepsy were the most important factors among the ones studied that were associated with social functioning and communication. We concluded that despite the heterogeneity of CP, only a small number of factors could explain the majority of the variance of functioning on distinct domains of activities and participation.

Chapter 3 presents the longitudinal course of motor functioning over two years in relation to associated factors at the level of impairments. The results showed that gross motor functioning, measured with the Gross Motor Function Measure-66 (GMFM-66), was stable over 2 years for the whole group. No differences were found in the course of the GMFM-66 scores between the different GMFCS levels. Within the GMFCS levels, significant differences were found in the course over two years for the different levels of limb distribution, selective motor control, muscle strength, range of motion in the hip and knee, spasticity of the hamstrings and type of education. The more severely affected children showed significantly larger decreases than the mildly affected children did. Multivariate analyses showed that poor selective motor control was the most important determinant of a less favourable course. We concluded that the above mentioned impairment characteristics, especially poor selective motor control, might be used to identify the children at risk for deterioration in gross motor function, and may serve as a guide for interventions.
In Chapter 4 we investigated the level and course of social functioning and communication of children with CP in comparison with typically developing children, and we determined the relation with disease characteristics, personal factors and environmental factors. The results showed that at baseline 45% of the children with CP had restrictions in social functioning and 74% had restrictions in communication. The course of social functioning showed an increase in restrictions in all children with CP, in comparison with data from the US general population. Restrictions in communication increased in children in GMFCS level V. Apart from disease characteristics (GMFCS level, epilepsy and speech problems), personal factors (externalizing behaviour problems) and environmental factors (having no siblings, low parental level of education and parental stress) were associated with more restrictions in social functioning and communication.

The results confirm that restrictions in social functioning and communication are an important issue for children with CP. Our conclusion was that it is important not only to focus on the medical treatment of children with CP, but also on their behavioural problems and social circumstances, and to support the parents in order to improve social functioning and communication in children with CP.

In Chapter 5 and 6 the subjective experience on functioning, global self-worth and perceived competence, and health-related quality of life were investigated. In Chapter 5 self-worth and perceived competence were examined in a subgroup within our study population: children who were able to complete self-report questionnaires. The findings of our sample were compared to the self-worth and perceived competence of Dutch age-mates from the general population. Furthermore we investigated the relationship with gross motor functioning and behaviour problems. The results showed that self-worth and perceived competence of the CP children were comparable to the Dutch norm sample. Only perceived athletic competence was rated lower than the norm sample. Within the CP sample, gross motor functioning was only associated with perceived motor competence. In the multivariate analyses internalising behaviour problems were associated with lower perceived competence on all domains and with lower global self-worth. In addition, results revealed a positive association between aggression and perceived motor competence, physical appearance and global self-worth, but only when gross motor functioning was included as associated variable.

We concluded that children with CP in general had a positive perceived competence and self-worth comparable to children without disabilities and trust themselves to meet the demands and expectations appropriate to their age and development. Low perceived competence on all domains and low self-worth were associated with internalising behaviour problems. Higher perceived competence on motor functioning, physical appearance and self-worth were associated with aggressive behaviour problems.

In Chapter 6 health-related quality of life (HRQOL) was investigated in a subgroup of our study population: children that were able to complete at least one domain of the TNO-AZL Children's Quality of Life Questionnaire (TACQOL). First, the findings of our sample at baseline, regarding children's HRQOL as reported by the children themselves as well as reported by their parents as proxy respondents, were compared to
the HRQOL of Dutch age-mates. Secondly, the child-reported HRQOL was compared with the parent-reported HRQOL of the children. Finally, we investigated the course of child-reported and parent-reported HRQOL over a three-year period, and investigated the relationship with gross motor functioning and behaviour problems. The results showed that both children with CP and their parents reported a significantly lower HRQOL in comparison with the normative sample. The exception was the domain of physical complaints, in which children with CP reported fewer complaints such as pain, dizziness. In comparison to their parents’ perceptions, the children reported a significantly better HRQOL. The longitudinal analyses showed that, according to the children, the HRQOL remained stable in all domains, except for autonomous functioning, in which an improvement was found. Parents reported an improvement for the domains autonomous and cognitive functioning and negative moods. The results showed a small but significant relationship between HRQOL and gross motor functioning and showed a strong relation between HRQOL and internalising behaviour problems (child and parent-reported HRQOL) and externalising behaviour problems (only parent-reported HRQOL).

We concluded that children with CP have a lower HRQOL than age-mates, and that the HRQOL is not only influenced by the motor functioning of the children, but even more by their behavioural problems. In addition, it should be noted that the negative HRQOL of children with CP is qualified by the person who reports: children with CP appeared as more positive than their parents.

In Chapter 7, the general discussion, we summarised and discussed the main findings of this thesis, the implications for clinical practise, some methodological considerations, and we addressed recommendations for future research. The results of the three studies on functioning showed considerable limitations in distinct domains of functioning. The longitudinal analyses showed deterioration of social functioning in all children with CP, and deterioration of motor functioning and communication in subgroups of children with CP. Professionals might use the identified determinants of deterioration to identify children at risk for a decrease in functioning. The results of the two studies on the subjective experiences showed that the subjective feelings of children with CP do not necessarily correspond to the objective level of functioning of children with CP. Behavioural problems proved to have a significant association with the level and course of functioning, self-esteem and HRQOL.

Recommendations for future research included longer follow-up studies to investigate the course of functioning into adolescence and early adulthood. Furthermore, the associated factors that were identified in our longitudinal analyses, should be used to develop intervention studies, in which should be investigated whether the identified relations are causal. Special attention should be paid to the behavioural problems in different age-groups of children with CP.