Health Issues, Activity And Participation Of Young Adults With Cerebral Palsy

Marloes van Gorp

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A long-term follow-up study

Marloes van Gorp

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General introduction

Individuals with cerebral palsy (CP) face challenges related to the childhood-onset disability throughout their lifespan. Therefore, health and functioning of these individuals is of interest not only in childhood, but remains so further on in their lives. Children and adults with CP often report health issues such as pain and fatigue, but knowledge on these issues in young adults is limited. Furthermore, previous studies have described that children and adults with CP experience activity limitations and participation restrictions. However, little is known of the development of activity and participation into and throughout adulthood. Better insight in development on the long-term is needed to guide treatment decisions and to adequately guide expectations of individuals with CP and their families on their future functioning in daily life. Furthermore, it is unknown if childhood factors can predict participation as an adult. If so, these factors determine which children with CP are at risk for restrictions in participation in adulthood and may serve as potential targets for treatment. This introductory chapter expands on the background of experienced health, activity and participation of young adults with CP, and concludes with the aims of this thesis.

Cerebral palsy (CP)

CP is the most frequent childhood-onset disability with a prevalence of 2 per 1000 live births in Europe.[5] CP is defined as 'a group of permanent disorders of the development of movement and posture, attributed to non-progressive disturbances that occurred in the developing fetal or infant brain, causing activity limitation'.[6] The motor disorders are often accompanied by other impairments, including epilepsy and disturbances of cognition, communication and behavior.[6,7]

To describe the wide variation in clinical features and functional abilities presented by individuals with CP, some characteristics are often used to classify these. The motor disorders can be classified according to subtype: spastic, ataxic or dyskinetic CP.[8] In addition, the severity of gross motor function limitations can be classified using the five-level Gross Motor Function Classification System (GMFCS). Figure 1 presents the five GMFCS levels, that range from walking without limitation (level I) to being severely limited in self-mobility and posture control (level V). Individuals with GMFCS level IV or V are unable to walk independently.

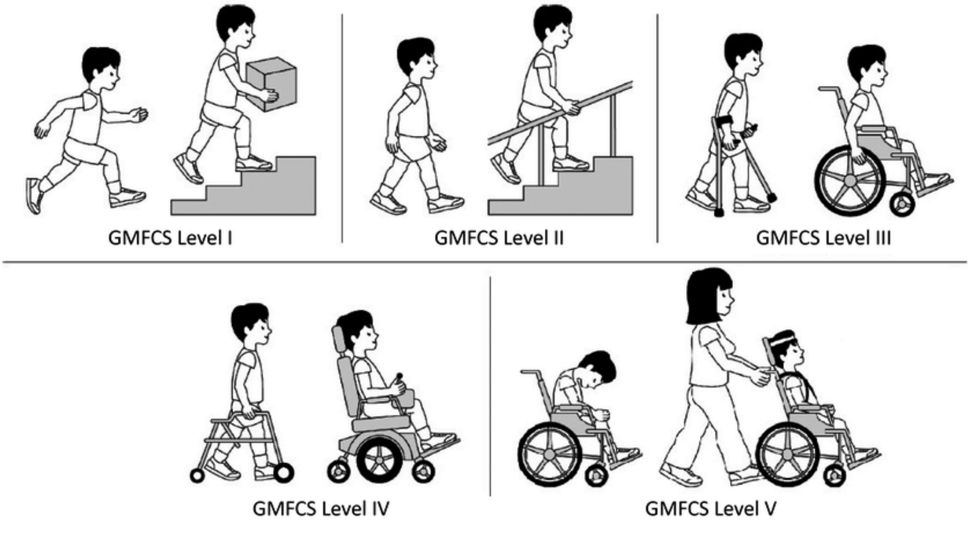

Figure 1: Flow chart of inclusion of study participants

Lifespan perspective in CP

In recent decades, a large majority of children with CP survives into adulthood.[9] Accordingly, nowadays most individuals with CP are adults.[10] Still, most CP research so far has focused on children; the number of studies focusing on adolescents transitioning to adulthood or adults with CP has only recently started to increase in quantity.[11]

The transition to adult life is a particularly demanding phase in the life course that comes with changes of desired or socially defined roles,[12] such as independent living, maintaining relationships and having a job. For individuals with CP, this transition into adulthood provides even more challenges than for their typically developing peers. This phase coincides with a decrease in utilization of rehabilitation services,[13] since pediatric care usually ends at the age of 18 and the transition from pediatric to adult care comes with difficulties.[14] Later in their lives, adults with CP often return to rehabilitation care with worsening health problems and participation restrictions.[15,16]

International Classification of Functioning, Disability and Health

In rehabilitation medicine, body functions and structures are primarily used to assess functioning and health of individuals with CP. Moreover, optimizing activity and participation is the main goal of rehabilitation treatment.[17,18] Body functions, activity and participation are often described using the universal language of the International Classification of Functioning, Disability and Health (ICF). Body functions are the physiological and psychological functions of the body, and include experienced health issues such as pain and fatigue. The ICF defines activity as 'the execution of a task or action by an individual' and participation as 'involvement in a life situation'. Furthermore, the ICF framework suggests that body functions and structure, activity and participation interact dynamically with an individual's health condition, environmental factors and personal factors (see Figure 2).[19]

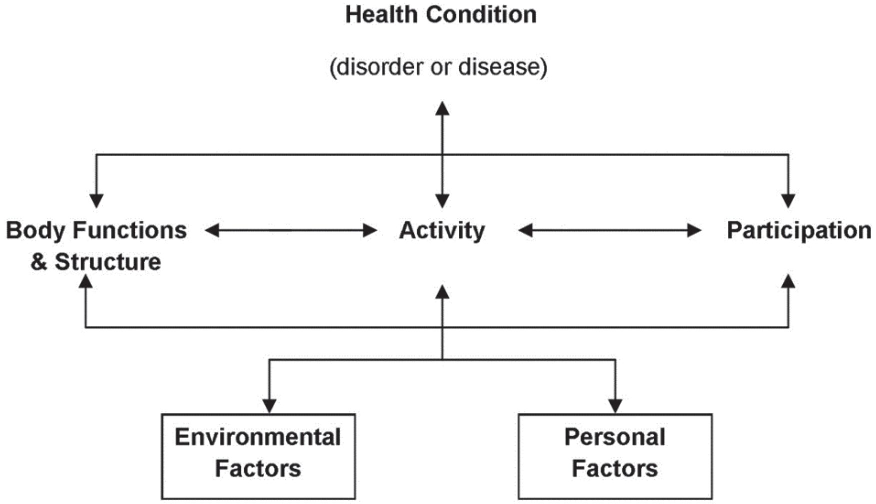

Figure 2: The framework of the international classification of functioning, disability and health International Classification of Functioning, Disability, and Health: ICF. Geneva: World Health Organization, 2001.

Body functions: health issues

Several health issues were previously described in studies of adults with CP. Pain and fatigue especially are often studied, found to be highly prevalent and found to occur more often than in the general population.[20-25] The literature is inconclusive on differences between subgroups of severity of motor impairment: some indicate more severely impaired individuals have more pain and fatigue,[20,26,27] while others did not confirm differences between subgroups.[25,28] Moreover, health issues in specific subgroups of individuals with CP were not compared to reference values. Therefore, studying pain and fatigue in individuals with CP in their twenties by level of motor functioning and in comparison to references could provide valuable insight in their health.

Pain and fatigue are known to be associated with depression in individuals with chronic neurological conditions such as stroke and traumatic brain injury.[29,30] Co-occurrence of pain, fatigue, and depressive symptoms was also found in adults with CP, and seemed higher compared to the general Dutch population.[25] Apart from the latter study, depression has been studied scarcely in individuals with CP. Sleep disturbances are more prevalent in children and youth with CP than in typically developing children.[31,32] Sleep disturbances are also known to relate to other health issues in children with CP and adults with other neurological disorders.[33,34] To our knowledge, sleep disturbances have not yet been studied in adults with CP. Knowledge on depressive symptoms and sleep disturbance and the clustering of health issues may establish the relevance of these health issues in rehabilitation of young adults with CP.

PERRIN study

The studies presented in this thesis are part of the prospective longitudinal cohort study Pediatric Rehabilitation Research in the Netherlands (PERRIN). The PERRIN study was explicitly set up to investigate the longitudinal development and determinants of activities and participation of individuals with CP. The program is a collaboration of several university medical centers and rehabilitation centers in the Netherlands. Participants in four age cohorts (baseline age: 1-2 years, 5-7 years, 9-13 years and 16-20 years) were measured three or four times between 2000 and 2007.1-4

Activity and Participation

The ICF distinguishes different domains of activity and participation, including: communication, mobility, self-care, domestic life, interpersonal interactions and relationships, major life areas (referring to education, employment and economic life) and community, social and civic life. Activity and participation in these domains can be qualified as *capacity* (what one can do in a standardized environment) or as *performance* (what one actually does do in their usual environment). Performance can be assessed as whether or not activities in participation domains are performed. In addition, specific aspects of performance in activity and participation can be assessed, such as whether individuals experience difficulty in performance.[19] Another aspect of participation is autonomy, referring to an individual’s freedom of decision and ability to act based on own attitudes and reasoning.[18] Autonomy in participation is specifically relevant for individuals transitioning to adult life.

Limitations in activity and participation

In previous studies, individuals with CP were found to be limited in activity and participation in childhood, adolescence and early adulthood.[1,35-39] The PERRIN study established that 20-30% of adolescents and young adults (16-20 years) with CP experienced difficulty in activity and participation.[1] Studies in adults with CP found that over 40% experienced difficulty in some domains of participation.[37,40] Regarding autonomy in participation, young adults with CP (age 20 years) less often live independently, have less experience with intimate relationships, and less often have a job, compared to their able-bodied peers.[41] However, it remains unknown how difficulty and autonomy in participation develop during their transition from adolescence into adulthood.

Associations with activity and participation

Previously, the PERRIN study and other literature described cross-sectional associations between participation and CP-related characteristics, body functions and environmental and personal factors in youth and young adults with CP.[1,4,42,43] These studies demonstrated that individuals who are more severely affected, those with intellectual disability, and those who have epilepsy are more restricted in their participation.[44] Additionally, some environmental (i.e. living environment, social environment) and personal factors (i.e. behavioral problems) were determinants of participation.[1,42,44] In addition to these cross-sectional associations, longitudinal studies are needed that provide information on factors predicting future participation, to enable clinicians to timely offer treatment.

Development of activity and participation in CP

Prognostic development curves of gross motor capacity have been determined for each level of GMFCS in a Canadian longitudinal cohort study.[45] These gross motor capacity curves are widely used in clinical practice and were validated for Dutch children in the PERRIN study.[46] They show that gross motor capacity development inclines fast up to 4 to 6 years and then levels off towards limits, that are distinctly lower for each level of GMFCS.

The PERRIN cohort studies also reported on development of performance of activity and participation. Data of the four cohorts were combined, and development of activity and participation was reported on between 2013 and 2016. The derived development curves described development of communication, motor performance, daily activities (including self-care, domestic and community life) and social interactions (including social life and interpersonal relationships and interactions) over the age range from 1 year to 24 years (and 1 to 16 years for those with intellectual disability). Development of expressive communication was primarily related to type of CP, while for receptive and written communication, development was more strongly associated to intellectual disability.[47] Development of motor performance was strongly related to level of GMFCS irrespective of ID, development of daily activities related to both level of GMFCS and intellectual disability, and social interaction related mainly to presence of intellectual disability.[48,49] At the end of the studied age range (22-24 yrs), performance of daily activities and social interaction seemed to still be increasing, so development had not yet stabilized.

Knowledge gaps

Although it has been demonstrated that adults with CP perceive problems with their health, presence of pain, fatigue, depression, sleep disturbances and the interrelation of health issues have not been reported thoroughly in young adults with CP. Neither was it studied how these health issues for subgroups of young adults with CP compare to the general population.

In the previous PERRIN cohort studies, development of performance of daily activities and social interaction seemed to be ongoing by the end of the age range; it needs yet to be confirmed at what age this development stabilizes for individuals with CP. Information on the long-term development of subdomains of activity and participation is therefore lacking. Additionally, development in communication was previously described for different subtypes of CP, but not for levels of GMFCS, which is of interest as well, as level of GMFCS and communication performance have recently been shown to be associated.[50,51]

Regarding other aspects of participation, i.e. difficulty and autonomy in participation performance, knowledge on the development in emerging adults with CP is lacking. Because of significant changes in life roles during the transition into adult life, insight in development of difficulty and autonomy in participation is of particular interest during this phase. Providing information on participation development for these aspects of participation performance could support rehabilitation practitioners in making treatment decisions and inform individuals with CP and their families on their expected future functioning.

Another knowledge gap exists concerning early predictors of participation. To identify subgroups at risk of limited participation and potential treatment targets, it is important to know which factors can determine future functioning. Until now, for individuals with CP, predictors have only been studied among youth and with a five year follow-up.[39,52] So far, it remains unknown whether (and which) childhood factors can predict participation in adulthood.

PERRIN Follow-up study

This thesis describes results of a 13-year follow-up of the oldest two PERRIN cohorts, now 21-34 years old. This long-term follow-up of the PERRIN cohort studies enables us to describe health issues of young adults with CP, to determine development of activity and participation into the twenties and early thirties of individuals with CP, and to identify childhood predictors of their young adult participation levels.

Aims

This thesis focuses on health issues, activity and participation of young adults with CP. First it aims to describe health issues of young adults with CP compared to the general population of the same age. Second, it aims to extend the knowledge on development of performance in activity and participation of individuals with CP from childhood and adolescence into adulthood. Third, it aims to identify early predictors of young adult participation.

Outline of the thesis

The content of the chapters of this thesis is visually presented in Figure 3. Chapter 2 investigates the health issues pain, fatigue, depressive symptoms and sleep disturbance in young adults with CP in comparison to typically developing peers. Development of activity and participation performance from infancy into adulthood is described in Chapters 3 and 4. Chapter 3 presents development curves by level of GMFCS for motor performance and daily activities, separately for individuals with CP of average intelligence and those with intellectual disability. Chapter 4 presents development curves of communication and social interactions by level of GMFCS, separately for individuals with CP of average intelligence and those with intellectual disability. Chapters 5 and 6 describe development of difficulty (Chapter 5) and autonomy (Chapter 6) in participation in several life areas, among which domestic life, interpersonal relationships and employment of individuals with CP of average intelligence from adolescence into their twenties and early thirties. In Chapter 7 and 8 we identified which CP-related factors, aspects of

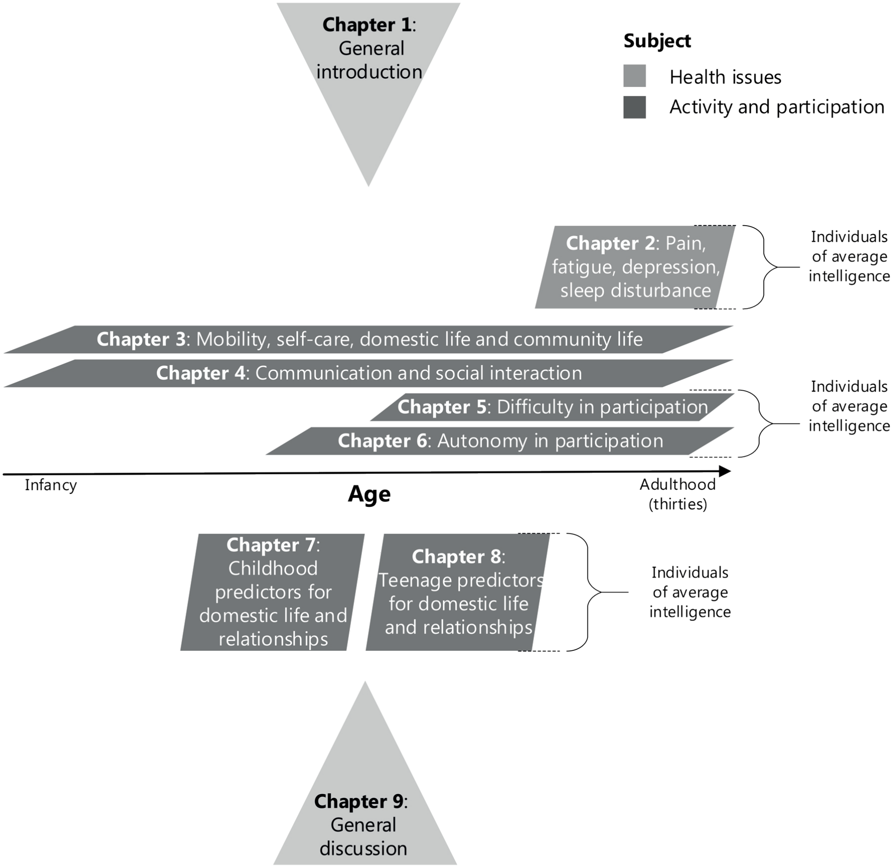

Figure 3: Chapter layout of 'Health issues, activity and participation of young adults with cerebral palsy'

Note: Chapters including only individuals of average intelligence are indicated. Other chapters (chapter 3, 4, and 7) include the full population of individuals with CP (i.e. also those with intellectual disability)

functioning, or environmental and personal factors assessed in children (Chapter 7) or teens with CP (Chapter 8) predict their adult participation in domestic life and interpersonal relationships. Finally, Chapter 9 discusses the main results of the thesis, and describes recommendations for future research and clinical implications.

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2

Pain, fatigue, depressive symptoms and sleep disturbance in young adults with cerebral palsy

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ABSTRACT

Objective: To investigate the occurrence and interrelatedness of pain, fatigue, depressive symptoms and sleep disturbance in young adults with cerebral palsy (CP) with different levels of Gross Motor Function Classification System (GMFCS) and their association with global health compared to age-matched references.

Design: Multicenter cohort study

Setting: Outpatient rehabilitation clinics of two university medical centers.

Participants: 97 individuals with CP (GMFCS I-V) aged 21-34 years without intellectual disability and 190 age-matched references.

Interventions: Not applicable.

Main Outcome Measures: Pain was assessed using the PROMIS® (Patient-Reported Outcomes Measurement Information System) numeric rating scale. Fatigue, depressive symptoms, sleep disturbance, global physical and mental health were assessed using PROMIS short forms. Scores were compared between individuals with CP and references using linear or logistic regression analyses. Correlation coefficients and linear regression analyses assessed interrelationships of health issues and its associations with physical and mental health .

Results: Individuals with GMFCS I had less pain, fatigue and depressive symptoms, while individuals with GMFCS II-V had more pain (54%, $p<0.001$) and those with GMFCS III-V more fatigue (39%, $p=0.035$) than references (pain: 26%, fatigue: 14%). In individuals with CP compared to references, pain and fatigue were more interrelated (correlation coefficients: 0.71 vs 0.41) and stronger associated with global mental health (standardized betas: -0.84 vs -0.25).

Conclusions: Young adults with GMFCS II-V report much higher levels of pain and those with GMFCS III-V report higher levels of fatigue than references. Pain and fatigue are highly interrelated and specifically relate to mental health in young adults with CP. Depressive symptoms and sleep disturbance seem to be no prominent health issues in young adults with CP.

INTRODUCTION

Cerebral Palsy (CP) is the most common childhood-onset physical disability.[1] Much of the past research on CP focused on children and physical problems, but in recent years attention has broadened towards functioning and health of adults with CP.[2] However, a knowledge gap exists on health issues of adults with CP, and young adults in their twenties in particular. During this phase, individuals grow into adult roles, including managing their health and healthcare.[3] Therefore, in order to identify possible healthcare needs, insight should be attained in health issues in young adults with CP.

Pain and fatigue are often studied, found to be highly prevalent and to occur more often than in the general population in adults with CP.[2,4-8] The literature is inconclusive on differences between subgroups of severity of motor impairment: some found no differences in subgroups,[8,9] while others indicated that more severely impaired individuals have more pain and fatigue.[6,10,11] Moreover, for specific subgroups of individuals with CP health issues were not compared to reference values. Therefore, studying pain and fatigue in individuals with CP in their twenties by level of motor functioning and in comparison to a reference population could provide valuable new insight.

Depressive symptoms and sleep disturbance have been studied less often in individuals with CP, but may be relevant issues as well and are assumed to correlate with pain and fatigue.[8,12] There is some evidence that adults with CP are at increased risk of depressive symptoms compared to the general population.[13] Also, depressive symptoms were more prevalent in a sample of adults with CP.[8] In this sample, pain, depression and fatigue were described as a symptom cluster, similar to the clustering that is observed in individuals with other chronic conditions.[8,14,15] Furthermore, sleep disturbance is recognized as an underemphasized health issue in individuals with disabilities and occurs frequently in children with CP,[16,17] but was not studied in adults yet. Knowledge on depressive symptoms and sleep disturbance and the clustering of health issues may establish the relevance of these health issues in treatment of young adults with CP.

Individuals' overall health and wellbeing is reflected in their perceived global health. Global health is known to be associated with pain and fatigue in adults with CP,[7,18,19] but the association with depressive symptoms and sleep disturbance is unknown. Moreover, it is unknown whether associations between health issues and global health differ between individuals with CP and the general population. Insight in these associations may inform health professionals on the impact of health issues and can help to prioritize treatment goals.

To gain insight in the severity and impact of health issues in young adults with CP, we aimed to assess the level of pain, fatigue, depressive symptoms and sleep disturbance in young adults with CP of subgroups of GMFCS level compared to the general population of the same age. Second, we aimed to study how these health issues interrelate. Additionally, we aimed to determine their association to global physical and mental health.

METHODS

Participants

This study is part of the PERRIN (Pediatric Rehabilitation Research In the Netherlands) program that recruited cohorts of children with CP between 2002 and 2007 in the Netherlands.

Participants of the previous PERRIN 9-16 and PERRIN 16-24 cohort studies were invited to participate in a 13-year follow-up (PERRIN DECADE, current age: 21-34 years). The recruitment process of the cohorts was described in detail elsewhere.[3,20] Exclusion criteria for the present study were additional disorders affecting motor functioning or having an intellectual disability, which was classified as having attended special education for children with intellectual disability.[20] Of all 211 participants of the PERRIN 9-16 and PERRIN 16-24 cohorts, 176 participants were eligible and 167 were invited to participate (Figure 1). The study was approved by the medical ethical committees of VU University Medical Center and Erasmus MC University Medical Center.

Measurements

CP characteristics were classified by a trained physical or occupational therapist. Health issues and global health were assessed using self-report questionnaires in an online survey.[21]

CP characteristics

Type of CP was classified as unilateral spastic CP, bilateral spastic CP or non-spastic CP.[22] The level of functional ability was classified using the Gross Motor Function Classification System (GMFCS), Manual Ability Classification System (MACS) and Communication Function Classification System (CFCS), ranging from I (most able) to V (least able).[23-25]

Health issues and global health

The average pain severity over the past week was assessed using an 11-point numeric rating scale ranging from 0 (no pain) to 10 (worst pain imaginable), which is one of the items of the PROMIS® (Patient-Reported Outcomes Measurement Information System) Global Health Version 1.2 scale (Global07r).[26] This item was found valid and reliable to assess pain intensity in adults with CP.[27] Fatigue, depressive symptoms and sleep disturbance were assessed using the Dutch-Flemish translations of the PROMIS v1.0 short forms 8a.[26,28-31] These PROMIS short forms consist of 8 items that are 7-day recall statements, scored on 5-point Likert scales. Physical and mental health were assessed using the PROMIS Global Health Version 1.2 scale.[32] Psychometric properties of PROMIS measures for Fatigue, Depression, Sleep Disturbance and Global Health have been studied extensively and show acceptable to excellent reliability and validity across several United States (US) and Dutch (clinical) populations.[30,31,33-37]

Reference data

From two samples representative of the Dutch population, collected for validation studies of the Dutch-Flemish PROMIS instruments, observations of individuals aged 21-34 years were selected (C. Terwee, personal communication, September 17, 2018).[26] The samples were composed to be representative of the general population with a maximum of 2.5% deviation from the distributions of gender, age, education, ethnicity and region, based on data from Statistics Netherlands in 2016 or 2013 (depression sample). The Fatigue (95 items) and Sleep Disturbance item banks (27 items) and Global Health Version 1.2 scale (including the pain item) were answered by one sample (n=190 aged 21-34), while the Depression item bank (28 items) was answered by another sample (n=202 aged 21-34).

PROMIS T-scores

Response pattern scoring was used to transform the PROMIS short form (adults with CP) or full item banks (reference populations) and global health scale scores into a T-score.[34,37]

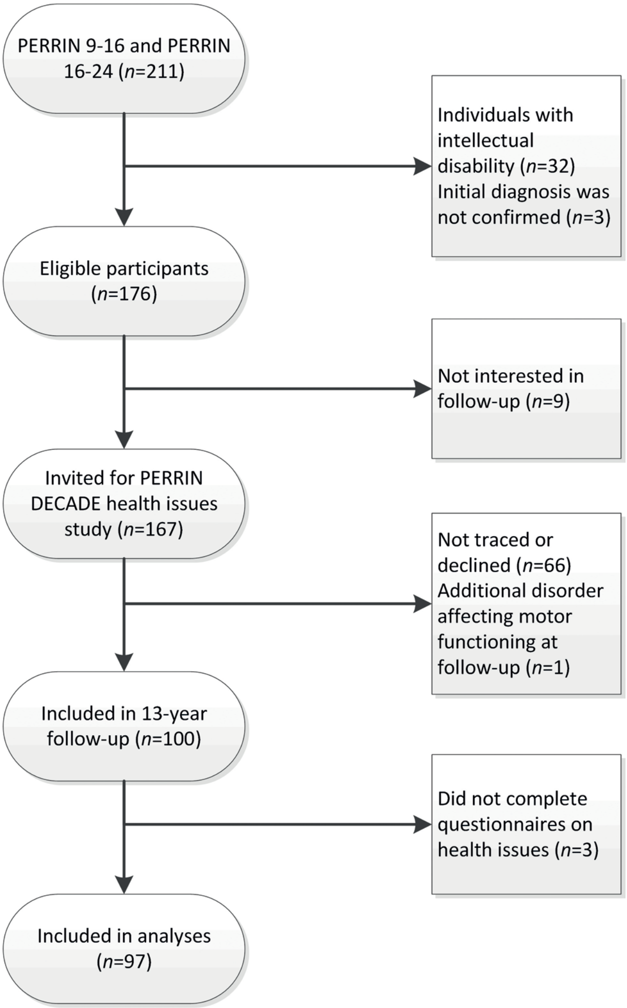

Figure 1: Flow chart of inclusion of study participants

A T-score of 50 (with a SD of 10) represents the average of the US centering sample; a representative sample from the US general population. Higher scores indicate more of the concept being measured (i.e. more fatigue, better physical health).

Cut-offs

Pain scores 1-3 were considered mild, scores 4-7 moderate and scores 8-10 severe.[38] In line with the PROMIS guidelines, scores for fatigue, depressive symptoms, and sleep disturbance of 55-60 were considered mild issues, 60-70 moderate issues and >70 severe issues.[39] Global mental and physical health scores were classified 'poor' (<34 for physical, <28 for mental health) to 'excellent' (>58 for physical, >57 for mental health), based on US population estimates.[39,40]

Statistical analysis

SPSS for Windows was used for analyses and a p-value <0.05 was considered statistically significant (SPSS 22, IBM SPSS Statistics, Armonk, NY, USA). To study the potential influence of loss to follow-up since baseline, the distribution of gender, GMFCS level and type of CP of individuals who were lost to follow-up (*n*=79) were compared to those included (*n*=97) using a chi-square test, with Cramer's V for effect size. The median and interquartile range of pain severity, and means and standard deviations fatigue, depressive symptoms and sleep disturbance, physical health and mental health were computed for the total CP sample, GMFCS subgroups and reference samples. Additionally, frequencies of mild, moderate and severe issues were calculated. To study differences between groups, scores of individuals with CP (total sample and GMFCS subgroups I, II and III-V) and reference groups were compared using logistic regression for pain (scores >3) and linear regression analyses for fatigue, depressive symptoms and sleep disturbance. These analyses were corrected for gender, since it was assumed to be a confounder.[4,6]

To study clustering of health issues, associations were determined between pain, fatigue, depressive symptoms and sleep disturbance for individuals with CP and references using Spearman (for associations with pain) and Pearson correlation coefficients, except for associations with depressive symptoms in references because these were assessed in another sample. The 95% confidence intervals (CIs) of the estimated correlation coefficients indicated whether correlation coefficients were significantly different between groups.

To study associations of pain, fatigue, depressive symptoms and sleep disturbance with global physical and mental health, linear regression analyses were conducted separately for individuals with CP and references (except for depressive symptoms in the reference population). Then, analyses were performed on the combined data of both groups, including a group*health issue interaction for pain, fatigue, and sleep disturbance, to determine whether associations differed between individuals with CP and the general population.

RESULTS

Participant characteristics are presented in Table 1; 97 young adults with CP without ID completed the measurements (58% response rate, mean age: 28.5y [SD: 3.8y], 59% male). Loss to follow-up was not related to gender, GMFCS or CP-type (p-values: >0.65, Cramer's Vs: 0.02, 0.09 and 0.12, respectively). Individuals in the reference sample for pain, fatigue and sleep disturbance (n=190) had a mean age of 28.1y (SD: 3.7y) and 56 (30%) were male, for depressive symptoms (n=202) they had a mean age of 28.1y (3.9y) and 76 (38%) were male.

No differences between individuals with CP and references were found in moderate to severe pain occurrence (NRS>3), and severity of fatigue and sleep disturbance. Depressive symptoms were less severe in individuals with CP (Table 2, Table 3). However, when individuals with GMFCS level I, level II and level III-V were analyzed separately, those with GMFCS level I had less severe pain, fatigue and depressive symptoms compared to references. Those with GMFCS level II and III-V on the other hand had higher odds of pain (OR [CI]: 3.84, [1.69-8.74] and 4.32 [1.55-12.02]) and those with GMFCS level III-V had somewhat more severe fatigue (beta [SE]: 4.61 [2.31]) compared to references (Table 3). Figure 2a shows the occurrence of mild, moderate and severe health issues in the GMFCS subgroups and references. Of those with GMFCS level II-V 54% reported pain and of those with GMFCS level III-V 39% reported fatigue. In the reference samples 26% reported pain and 15% reported fatigue.

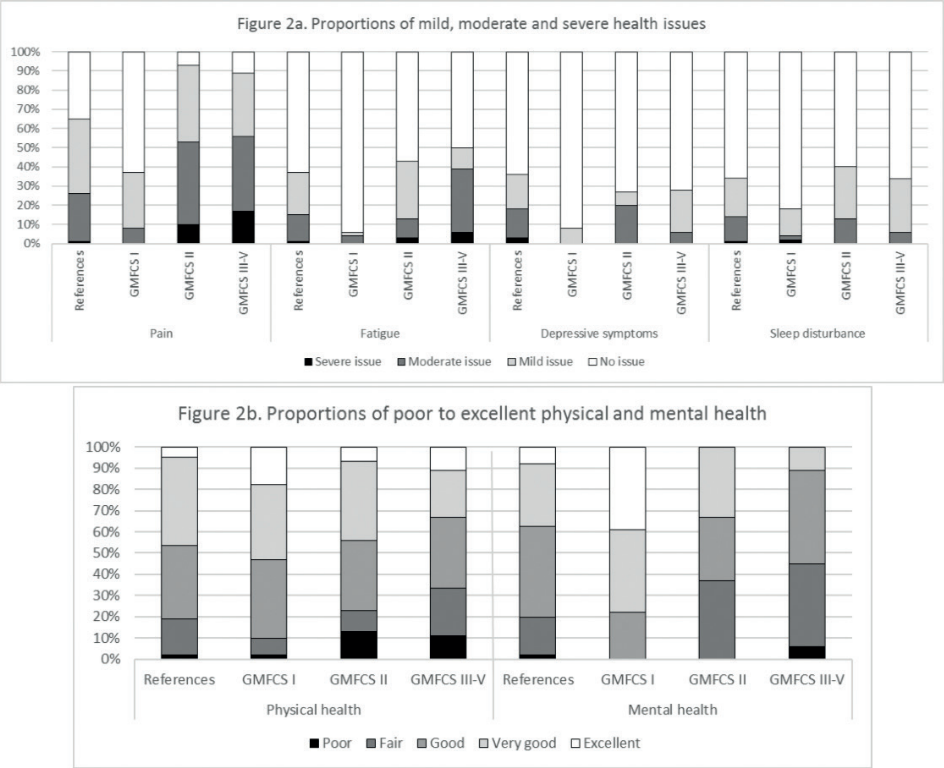

Figure 2: proportions of health issues (a) and physical and mental health (b) of references (*n*=190 for pain, fatigue, sleep disturbances and global health, *n*=202 for depressive symptoms), individuals with GMFCS level I (*n*=49), GMFCS level II (*n*=30) and GMFCS level III-V (*n*=18)

There were positive associations between all health issues in both the individuals with CP and references (see Figure 3). Pain and fatigue correlated more strongly in individuals with CP compared to references (r=.71 vs r=.41), and fatigue and sleep disturbance correlated more weakly (near-significant, r=.57 vs r=.70).

All health issues related negatively to global physical and mental health in individuals with CP and references with standardized betas ranging from -0.25 to -0.84. In individuals with CP pain and fatigue had a stronger association with mental health, and a weaker association with physical health compared to references, as indicated by significant interactions (see Table 4).

DISCUSSION

This study provided insight in health issues in individuals with CP in their twenties. Young adults with CP with GMFCS level II-V more often reported moderate to severe pain (54%) and those with GMFCS level III-V more often reported moderate to severe pain fatigue (39%) than a reference group (pain: 26%, fatigue: 15%). In contrast, those with GMFCS level I had lower levels of pain, fatigue and depressive symptoms. Pain and fatigue showed a stronger interrelationship in individuals with CP, and correlated more strongly to mental health compared to a reference group.

Table 1: Characteristics of study participants

	Participants with CP without intellectual disability (*n* = 97)
Mean age (SD); age-range	28.5y (3.8y); 21.6y-34.3y
Male/female, *n* (%)	59/38 (61/39)
CP type, *n* (%)	
- Unilateral spastic CP	39 (40)
- Bilateral spastic CP	44 (45)
- Non-spastic CP	14 (15)
GMFCS level, *n* (%)	
- I	49 (51)
- II	30 (31)
- III	5 (5)
- IV	10 (10)
- V	3 (3)
MACS level, *n* (%)	
- I	49 (51)
- II	41 (42)
- III	4 (4)
- IV	3 (3)
- V	-
CFCS level, *n* (%)	
- I	78 (80)
- II	17 (18)
- III	2 (2)
- IV	-
- V	-

SD: standard deviation
CP: cerebral palsy
GMFCS: Gross Motor Function Classification System
MACS: Manual Ability Classification System
CFCS: Communication Function Classification System

Pain in subgroups of GMFCS levels had not been compared to reference populations before. Our result of much higher moderate to severe pain occurrence in individuals with GMFCS level II-V compared to GMFCS level I is in line with the meta-analysis that found more pain in individuals with CP with GMFCS level II and IV compared to GMFCS level I.[6] Some other studies did not identify this difference, which may be explained by lower numbers of individuals with GMFCS level I in these populations, and therefore they may not have had enough power to determine the difference.[8,9] Furthermore, these populations had a slightly higher age which may explain the different results. In conclusion, pain occurs often in young adults with GMFCS level II-V and we therefore advice professionals to monitor pain in these individuals.

Table 2: Descriptives of perceived health outcomes of young adults with CP without intellectual disability and an age-matched reference population

	CP (all)	GMFCS level I	GMFCS level II	GMFCS level III-V	Reference population
Health issues	**mean (SD); *n*=97**	**mean (SD); *n*=49**	**mean (SD); *n*=30**	**mean (SD); *n*=18**	**mean (SD); *n*=190/202**[a]
Pain[b], NRS-scale, range 0-10	2 (0-4.5)	0 (0-2)	4 (2-6)	5 (2-7)	1 (0-4)
Fatigue, PROMIS, T-score	48.6 (9.9)	44.3 (8.0)	52.2 (8.2)	54.3 (12.3)	50.5 (9.5)
Depression, PROMIS, T-score	46.7 (8.6)	43.9 (6.7)	49.5 (10.6)	49.4 (7.3)	51.5 (9.4)
Sleep disturbance, PROMIS, T-score	50.1 (7.8)	48.1 (8.0)	51.8 (8.0)	52.8 (5.8)	50.9 (8.3)
Global health	**mean (SD); *n*=97**	**mean (SD); *n*=49**	**mean (SD); *n*=30**	**mean (SD); *n*=18**	**mean (SD); *n*=190**
Physical health, PROMIS, T-score	49.0 (8.6)	51.6 (7.7)	46.4 (9.0)	46.0 (8.7)	48.7 (7.1)
Mental health, PROMIS, T-score	47.9 (9.9)	54.3 (7.3)	42.5 (7.3)	39.3 (8.1)	45.7 (7.9)

[a] Reference population for depression, n=202
[b] Median (interquartile range)
CP: cerebral palsy
SD: standard deviation
GMFCS: Gross Motor Function Classification System
NRS: Numeric Rating Scale
PROMIS: Patient-Reported Outcomes Measurement Information System

Regarding fatigue, we found somewhat higher severity levels in individuals with CP with GMFCS level III-V (mean T-score: 54.3) compared to those in the reference population (mean T-score: 50.5). Previous studies did not compare subgroups of GMFCS to reference values, but did find that individuals with lower motor function experience more fatigue compared to those with higher motor function.[8,10] Therefore, our results indicate that fatigue is an issue in individuals with GMFCS level III-V in their twenties and we suggest monitoring in rehabilitation.

Young adults with CP did not have higher levels of depressive symptoms and sleep disturbance compared to an age-matched reference group. This is in contrast with the literature that found individuals with CP at higher risk of depression compared to reference values.[8,12] A possible explanation for this may be a floor effect in the assessment of depressive symptoms. This floor effect was observed in other studies as well,[36] and is more pronounced in the short form that was used for individuals with CP, than in the item bank that was used in the reference population.[41] Because of this, the Depression short form does not seem to adequately cover low levels of depressive symptoms,[41] which may have led to an underestimation of the mean score of individuals with CP. Still, this only affects the scores of those with low levels of depressive symptoms, the scores of individuals with moderate to severe depressive symptoms were not affected. Therefore, since depressive symptoms and sleep disturbance are not severe in individuals with CP in their twenties they may not require specific attention in rehabilitation care.

Table 3: Regression coefficients for the differences in pain, fatigue, depression and sleep disturbance between individuals with CP and references

	Dependent	**Pain**	**Fatigue**	**Depressive symptoms**	**Sleep disturbance**
	Independent	Exp(B); 95% CI; p-value	beta (SE); std beta; p-value	beta (SE); std beta; p-value	beta (SE); std beta; p-value
Crude analysis	**CP (versus references)**	1.25; 0.73 to 2.15; p=0.410	-1.87 (1.20); -0.09; p=0.121	-4.78 (1.14); -0.24; p<0.001	-0.78 (1.02); -0.05; p=0.448
Adjusted for gender	**CP (versus references)**	1.50; 0.85 to 2.67; p=0.163	-1.03 (1.25); -0.051; p=0.412	-4.87 (1.11); -0.24; p<0.001	-0.35 (1.07); -0.02; p=0.742
	Dependent	**Pain**	**Fatigue**	**Depressive symptoms**	**Sleep disturbance**
	Independent	Exp(B); 95% CI; p-value	beta (SE); std beta; p-value	beta (SE); std beta; p-value	beta (SE); std beta; p-value
Crude analysis	**GMFCS I (versus references)**	0.25; 0.09 to 0.73; p=0.011	-6.17 (1.49); -0.24; p<0.001	-7.51 (1.45); -0.30; p<0.001	-2.78 (1.30); -0.13; p=0.033
	GMFCS II (versus references)	3.20; 1.46 to 7.03; p=0.004	1.74 (1.83); 0.06; p=0.343	-1.97 (1.78); -0.06; p=0.268	0.89 (1.59); 0.03; p=0.578
	GMFCS III-V (versus references)	3.50; 1.31 to 9.37; p=0.013	3.81 (2.30); 0.10; p=0.098	-2.05 (2.23); -0.05; p=0.360	1.92 (2.00); 0.06; p=0.331
Adjusted for gender	**GMFCS I (versus references)**	0.30; 0.10 to 0.89; p=0.030	-5.32 (1.54); -0.21; p=0.001	-7.62 (1.48); -0.30; p<0.001	-2.35 (1.34); -0.11; p=0.081
	GMFCS II (versus references)	3.84; 1.69 to 8.74; p=0.001	2.43 (1.85); 0.08; p=0.190	-2.06 (1.79); -0.07; p=0.252	1.24 (1.62); 0.05; p=0.445
	GMFCS III-V (versus references)	4.32; 1.55 to 12.02; p=0.005	4.61 (2.31); 0.12; p=0.047	-2.15 (2.25); -0.06; p=0.339	2.33 (2.03); 0.07; p=0.252

CP: Cerebral Palsy
GMFCS: Gross Motor Function Classification System
Exp(B): odds ratio
SE: standard error
std beta: standardized beta

Remarkably, we did not find a difference in pain and fatigue between the total sample and the reference population, contrasting other studies.[4,5,8,42] This can be explained by the large proportion of individuals with GMFCS level I in our sample, that had very low levels of pain and fatigue. These low levels may be related to the lifelong aspect of their disability. Similar to the response shift theory, individuals with CP are accustomed to functioning with their disability, may have struggled with health issues in the past, and therefore rate their health more positively.[43]

Our results support the idea that there is a clustering of pain and fatigue in individuals with CP,[5,44] which was also found in individuals with stroke.[14] Because of the low occurrence of depressive symptoms we did not confirm a triad of pain, fatigue and depressive symptoms that was previously found in a population of individuals with bilateral CP and in individuals after stroke.[8,14] Furthermore, the association between fatigue and sleep disturbance tended to be less prominent in individuals with CP compared to references. This may be caused by a stronger correlation in individuals with CP between fatigue and other health complaints, like pain or physical limitations, than between fatigue and sleep. For the healthier reference group on the other hand, sleep problems could be a major issue causing fatigue. Although the underlying mechanism of the association between pain and fatigue is unclear, health professionals may consider treating pain and fatigue in combination if both issues occur. A lifestyle intervention program may for instance be fitting, since this was previously found to reduce both fatigue and pain.[45]

All health issues had a negative association with global health. This is in line with previous studies that found associations between pain and fatigue with global health in children with CP and associations of pain and fatigue with health concerns of adults with CP.[18,19] In addition, pain and fatigue were more strongly associated to mental health, but weaker to physical health in young adults with CP compared to the general population. Pain and fatigue therefore seem to affect mood and quality of life, which determine the PROMIS global mental health score. Global physical health was less affected by pain and fatigue in individuals with CP compared to the reference population in our study. This may be explained by their physical disabilities, that may be dominant in rating their global physical health. In conclusion, our results suggest that pain and fatigue should be intervened on timely in young adults with CP to prevent reduced mental health. In addition, in line with a recently developed patient-centered research agenda for CP, future research may aim to determine which variables, in addition to GMFCS level predict the occurrence of pain and fatigue in young adulthood.[46]

Strengths and limitations

Strengths of this study are studying four health issues and global health simultaneously, thus allowing to disentangle associations between them and directly compared the results to a reference population of the same age. Limitations mainly concern representativeness of the used samples. First, although loss to follow-up was not selective regarding gender, GMFCS level and type of CP, and the distribution of these factors was similar to population-based studies when excluding those with intellectual disability,[47] we cannot rule out that other factors may have caused individuals to drop out from the study. Second, the reference groups were representative of the Dutch population within the age range 18-40 years.[26] Since we selected a subgroup from this, deviations from norm data distributions may exceed 2.5%, as was observed for gender. In addition, we combined subgroups of individuals with GMFCS levels III-V, because we had low numbers of individuals with GMFCS levels III and V.

Table 4: Regression coefficients for the relations between pain, fatigue, depression, sleep disturbance and physical and mental health for individuals with CP and references*

Dependent	**Physical health**			**Mental health**		
	beta (SE); std beta; p-value			beta (SE); std beta; p-value		
Independent	CP	references	interaction	CP	references	interaction
Pain	-1.05 (0.31); -0.33; p=0.001	-1.98 (0.17); -0.64; p<0.001	0.93 (0.32); 0.24; p=0.004	-3.04 (0.21); -0.84; p<0.001	-0.86 (0.24); -0.25; p<0.001	-2.18 (0.34); -0.50; p<0.001
Fatigue	-0.34 (0.08); -0.39; p<0.001	-0.49 (0.04); -0.65; p<0.001	0.15 (0.08); 0.47; p=0.067	-0.73 (0.07); -0.73; p<0.001	-0.44 (0.05); -0.53; p<0.001	-0.29 (0.09); -0.78; p=0.001
Depressive symptoms	-0.63 (0.08); -0.63; p<0.001			-0.52 (0.11); -0.45; p<0.001		
Sleep disturbance	-0.44 (0.10); -0.40; p<0.001	-0.48 (0.05); -0.57; p<0.001	0.04 (0.10); 0.14; p=0.676	-0.60 (0.12); -0.47; p<0.001	-0.42 (0.06); -0.44; p<0.001	-0.18 (0.12); -0.51; p=0.130

CP: cerebral palsy
SE: standard error
std beta: standardized beta
*Correcting analyses for gender provides very similar results and are therefore not presented

CONCLUSIONS

Young adults with CP and GMFCS level II-V report higher levels of pain and those with GMFCS level III-V report higher levels of fatigue compared to age-matched individuals from the general population, while those with GMFCS level I report lower levels of pain, fatigue and depressive symptoms. Pain and fatigue are strongly interrelated and are specifically associated with mental health in young adults with CP. Therefore, we recommend that health professionals monitor pain and fatigue in young adults with GMFCS II-V and consider combined treatment for both health issues. Depressive symptoms and sleep disturbance seem to be no prominent health issues in individuals with CP in their twenties.

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3

Activity Performance Curves of Individuals with Cerebral Palsy

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ABSTRACT

Objective: To describe development curves of motor and daily activity performance from childhood into adulthood of individuals with cerebral palsy (CP) by functional severity level.

Methods: Participants with CP aged 1-20 years at baseline (n=421), Gross Motor Functioning Classification System (GMFCS) levels I-V (27% with intellectual disability [ID]), were longitudinally assessed up to 13-year follow-up. Motor and daily activity performance were assessed using the motor skills and daily living skills subdomains of the Vineland Adaptive Behavior Scales. Non-linear mixed effects analyses were used to describe development curves, estimating the limit (average maximal performance level) and age_{90} (age reaching 90% of the limit).

Results: Limits of gross and fine motor performance decreased with each lower functional level. Age_{90} was reached around 6-8 years in children with GMFCS or Manual Ability Classification System levels I-III, while those with lower functional levels approached their (lower) motor performance limits at a younger age. Limits of daily activity performance did not differ between individuals with GMFCS level I-III without ID. The age_{90}s of daily activity performance were reached between 11-14 years (personal), 26-32 years (domestic) and 22-26 years (community). Individuals with ID reached lower daily activity performance limits and approached their limits earlier.

Conclusions: Individuals with CP continue to develop motor performance after gross motor capacity limits are reached. For individuals with CP without ID daily activities continue to develop into adolescence and their twenties. Severely affected individuals functionally have the least favourable development of motor performance and those with ID of daily activity performance.

INTRODUCTION

Cerebral palsy (CP) is a common cause of childhood-onset disability that addresses disorders of movement and posture attributed to non-progressive disturbances of the developing fetal or infant brain, causing activity limitation throughout the lifespan.[1] The International Classification of Functioning and Health (ICF) defines activity as 'the execution of a task or action' which can be qualified by capacity (what one can do in a standard environment) or performance (what one actually does in their usual environment).[2] Activity covers activities related to motor functioning (e.g. mobility) and to daily life situations (e.g. activities in self-care, domestic life and community life). Children with CP can be limited in activities in these domains; moreover, these limitations are greater for those with lower levels of gross motor function (classified by the Gross Motor Function Classification System, GMFCS) or manual ability (classified by the Manual Ability Classification System, MACS).[3-6]

Apart from the degree of activity limitation, it is also important to describe its development for individuals with CP. Knowledge on long-term development can inform treatment decisions related to health development and also guide the expectations of (younger) individuals with CP and their parents regarding their future functioning.[7] The development curves of gross motor capacity, as measured with the Gross Motor Function Measure (GMFM), are widely used and have lower limits for each lower level of GMFCS. These curves have been validated for Dutch individuals with CP in the PERRIN study (Pediatric Rehabilitation Research in the Netherlands).[8,9] Recently, development curves of fine motor capacity (as measured using the Assisting Hand Assessment) were estimated for MACS levels I-III, and showed development curves similar to those of gross motor capacity.[10] However, for gross and fine motor *performance* similar developmental data are not yet available. Although motor capacity and performance are related,[11-13] motor capacity development does not necessarily translate to identical motor performance development; indicating that the motor capacity curves do not provide information on performance.[8,14] The concept of performance is of additional interest, as it concerns activities in a person's usual environment, referring to what individuals actually do in their daily life.

Previously, the PERRIN study provided longitudinal trajectories of overall motor performance (up to age 16 years) and daily activity performance (up to age 24 years), measured using the Vineland Adaptive Behavior Scales (VABS). For individuals with mild physical impairment, development of motor and daily activity performance showed, on average, an incline up to values close to those of typically developing children. For children with GMFCS level IV-V motor performance limits were significantly lower, and for those with intellectual disability (ID) development of daily activity performance was less favourable.[15] Small numbers of observations at the end of the age range caused uncertainty about the limits of performance, and it was unknown whether development of performance had been completed by age 24 years.[15] In line with the emerging life course perspective of health development,[7,16] the development of performance further into adulthood should be considered.

Current knowledge on the development of motor and daily activity performance can be extended in several ways. First, the use of a method of analysis similar to that used in the creation of the gross motor capacity curves would provide more detailed information on the rate and average maximal performance level (limit) of development of motor and daily activity performance and allow comparison between motor capacity and performance curves.[9] Second, adding a 13-year follow-up measurement to the PERRIN study would enable more accurate estimation of the limits of performance and determine whether development of daily activity performance continues over age 24. Finally, discerning between specific domains of daily activities would be relevant, as development in different life areas occurs at different ages; development of self-care is expected to peak earlier than that of domestic or community activities.[17] In addition, the level of functional ability and ID may influence the performance development of these activities differently. Therefore, the present study aims to describe the development curves of gross and fine motor performance, as well as the performance of self-care, domestic and community activities of individuals with CP from childhood into adulthood based on level of functional ability.

METHODS

Participants

Participants of the PERRIN cohort study were longitudinally measured over 3 or 4 years between 2000 and 2007. The PERRIN study consisted of 421 participants in four age cohorts: PERRIN 0-5 (baseline age: 1-2 years, 4 annual measurements, *n*=97), PERRIN 5-9 (baseline age: 5 and 7 years, 3 annual measurements, *n*=116), PERRIN 9-16 (baseline age: 9, 11 and 13 years, 4 annual measurements, *n*=107) and PERRIN 16-24 (baseline age: 16-20 years, 3 biennial measurements, *n*=101; see Table 1). The recruitment process of the four age cohorts is published elsewhere.[11-13,18] Eligible participants had a clinical diagnosis of CP and they (or a proxy) had sufficient knowledge of the Dutch language to complete interviews. Participants or their caregivers gave informed consent. In the PERRIN 16-24 cohort, individuals with ID (IQ of approximately <70) were excluded.[18] A 13-year follow-up was performed in the 9-16 and 16-24 cohorts (*n*=121), indicated as the PERRIN DECADE study. Figure 1 is a flowchart of the inclusion of observations in the data-analysis. Appendix 1 shows the number of observations by cohort for all assessments. Approval was obtained from the Medical Ethics committees of the participating centers.

Measures

Motor and daily activity performance

Gross and fine motor performance and performance of personal, domestic and community daily activities were measured using the Dutch version of the VABS survey motor and daily living skills subdomains.[17,19] The VABS assesses performance by means of a semi-structured interview and the original version was found reliable and valid for typically developing individuals up to age 19 and adults with ID, the Dutch version was validated for individuals with ID.[17,20] Items addressed activities of daily life in gross (20 items) and fine motor performance (16 items), and personal (39 items, i.e. getting dressed, doing one's hair), domestic (21 items, i.e. doing laundry, cooking) and community daily activities (32 items, i.e. using bank account, job skills). These activities were scored as never performed (0), sometimes or partially performed (1), or usually or habitually performed (2). A validated Dutch version of the screener of the VABS was used for the PERRIN 0-5 cohort.[21] This screener contains less items for each subdomain and is age-appropriate for 0-12 years. As there were very few items for domestic and community daily activities, those scores of the PERRIN 0-5 cohort were discarded. Other VABS screener scores were linearly transformed to match the VABS survey scores. Gross and fine motor performance were not assessed in the PERRIN 16-24 cohort, since that study focused on daily activities and participation.[22] Consequently, gross and fine motor performance were described over an age range of 1-27 years, and daily activity performance (individuals without ID) over age ranges of 1-34 years (personal) and 5-34 years (domestic and community activities). Finally, due to the exclusion of individuals with ID in PERRIN 16-24, daily activity performance for this subgroup was described up to 27 years.

Characteristics of CP

Level of functional ability registered at baseline was described for gross motor functioning (GMFCS) and manual ability (MACS).[5,6] The GMFCS and MACS are classification systems based on functional abilities, ranging from I (highest level) to V (lowest level). GMFCS levels range from walking without limitation (level I) to being severely limited in self-mobility and posture control (level V); individuals with GMFCS level IV or V are unable to walk independently. MACS levels range from handling objects in everyday life easily (level I) to needing assistance in handling all objects or with simple actions (level V). Individuals were classified with ID if they had an IQ below 70, assessed by the Snijders-Oomen Nonverbal Intelligence Test (PERRIN 0-5 cohort),[23] the Raven's Colored Progressive Matrices (PERRIN 5-9 cohort),[24] or based on school type: those following a special education program for children with ID were classified as individuals with ID (PERRIN 9-16 cohort).[13]

Statistical analysis

Frequencies of gender, subtype of CP, GMFCS level, MACS level and ID were calculated, to describe baseline characteristics. To estimate the motor and daily activity performance development, similar to the gross motor capacity curves,[8,9] nonlinear mixed effects modelling was conducted on the five VABS subdomain scores for each GMFCS or MACS level separately for age (continuous variable), using R 3.2.5.[25] Gross motor performance was analysed using the GMFCS and fine motor performance using the MACS. To allow for comparison and ease of interpretation between the three domains of daily activity performance, those were all analysed using the GMFCS. In accordance with a previous PERRIN publication,[15] daily activity performance was analysed separately for individuals with and without ID. The nonlinear model has two parameters with straightforward clinical interpretations: the rate (speed of development) and limit (average maximal performance level for a subgroup). The used model assumes a limit of maximal potential performance, and a development that is rapid at first but levels off toward reaching this limit. These assumptions seem to fit the motor and daily activity performance development described by the reference values of the VABS.[17] To enhance interpretation, the rate parameters were used to calculate the average age by which individuals reached 90% of their limit (age_{90}). Higher values of age_{90} therefore indicate slower development toward the limit. The 95% confidence intervals (CIs) of the limit and age_{90} were calculated and used to detect differences between GMFCS or MACS levels, with significant differences if the 95% CIs did not overlap ($p<0.05$). The degree of inter-individual variability of the limit was estimated by including a random limit in the model. From the random variance around limits we calculated 50% ranges that encompass the estimated limits of 50% of the individuals in the analysis. Since development of the subdomains of motor and activity performance does not necessarily start at birth, a start age of modelled development was selected based on the best fit of all observations, irrespective of GMFCS or MACS level (according to the Akaike Information Criterion [AIC]). The residual standard deviations (SDs) of the models provide an indication of the model fit.

To allow for the low number of observations in certain subgroups according to functional level and ID, no daily activity performance curves were estimated for individuals with GMFCS level V without ID, and the observations of individuals with ID were combined for the daily activity performance curves of GMFCS levels I-III and GMFCS levels IV and V.

Table 1: Characteristics of the study population at baseline, and for subgroups at 13-year follow-up

	PERRIN 0-5	**PERRIN 5-9**	**PERRIN 9-16**		**PERRIN 16-24**	
	(*n*=97, observations: 314)	**(*n*=116, observations: 328)**	**(*n*=107, observations: 399)**	***13-year follow up (n=67*)***	**(*n*=101, observations: 260)**	***13-year follow-up (n=54*)***
Age at baseline or 13-year follow-up, mean (SD)	1y7m (0y2m)	6y3m (1y0m)	11y2m (1y8m)	*24y7m (1y7m)*	18y6m (1y6m)	*31y8m (1y5m)*
Sex, *n* (%)						
Males	56 (58%)	76 (65%)	67 (63%)	*45 (67%)*	60 (59%)	*29 (54%)*
Females	41 (42%)	40 (35%)	40 (37%)	*22 (33%)*	41 (41%)	*25 (46%)*
Subtype, *n* (%)						
Spastic	94 (97%)	98 (84%)	86 (80%)	*52 (77%)*	88 (88%)	*26 (85%)*
Unilateral	41 (42%)	42 (36%)	37 (35%)	*21 (31%)*	41 (41%)	*21 (39%)*
Bilateral	53 (55%)	56 (48%)	49 (46%)	*31 (46%)*	47 (47%)	*25 (46%)*
Dyskinetic	2 (2%)	14 (12%)	4 (4%)	*3 (5%)*	4 (4%)	*1 (2%)*
Ataxic	0 (0%)	4 (4%)	4 (4%)	*3 (5%)*	2 (2%)	*1 (2%)*
Mixed	1 (1%)	0 (0%)	14 (12%)	*9 (13%)*	7 (7%)	*6 (11%)*
Level of gross motor function, *n* (%)						
GMFCS level I	30 (31%)	56 (48%)	49 (46%)	*30 (45%)*	74 (73%)	*38 (70%)*
GMFCS level II	13 (13%)	20 (17%)	14 (13%)	*7 (10%)*	8 (8%)	*4 (7%)*
GMFCS level III	23 (24%)	17 (15%)	13 (12%)	*8 (12%)*	6 (6%)	*4 (7%)*
GMFCS level IV	21 (22%)	9 (8%)	13 (12%)	*9 (13%)*	12 (12%)	*7 (13%)*
GMFCS level V	10 (10%)	14 (12%)	18 (17%)	*13 (19%)*	1 (1%)	*1 (2%)*
Level of manual ability, *n* (%)						
MACS level I	24 (25%)	44 (38%)	39 (41%)	*26 (39%)*	69 (78%)	*41 (76%)*
MACS level II	39 (40%)	38 (33%)	33 (34%)	*22 (33%)*	15 (17%)	*10 (19%)*
MACS level III	15 (16%)	15 (13%)	9 (9%)	*7 (10%)*	2 (2%)	*1 (2%)*
MACS level IV	6 (6%)	8 (7%)	10 (10%)	*7 (10%)*	1 (1%)	*1 (2%)*
MACS level V	8 (8%)	11 (10%)	5 (5%)	*5 (7%)*	1 (1%)	*1 (2%)*
	Unknown: 5		Unknown: 11		Unknown: 13	
Intellectual disability, *n* (%)						
No	50 (52%)	79 (68%)	75 (70%)	*46 (69%)*	101 (100%)	*54 (100%)*
Yes	45 (46%)	35 (30%)	32 (30%)	*21 (31%)*	0 (0%)	*0 (0%)*
	Unknown: 2	Unknown:2				

*: 13-year follow-up n=121

RESULTS

Overall, 421 participants (50% GMFCS level I, 45% MACS level I, 73% without ID) contributed to 1428 VABS observations at ages 1-34 years. Table 1 presents the characteristics of the participants of the four age cohorts at baseline and for the two oldest cohorts at the 13-year follow-up. Drop-outs are described in Figure 1 and were not selective regarding sex or CP characteristics. The estimated average development curves by GMFCS or MACS level are shown in Figure 2. Parameter estimates (limit and age_{90}) and 50% ranges of the limits are reported in Table 2. The raw observations are shown in Appendix 2 with the estimated curves and 50% ranges of the limits for the motor and daily activity subdomains (Fig. A-E).

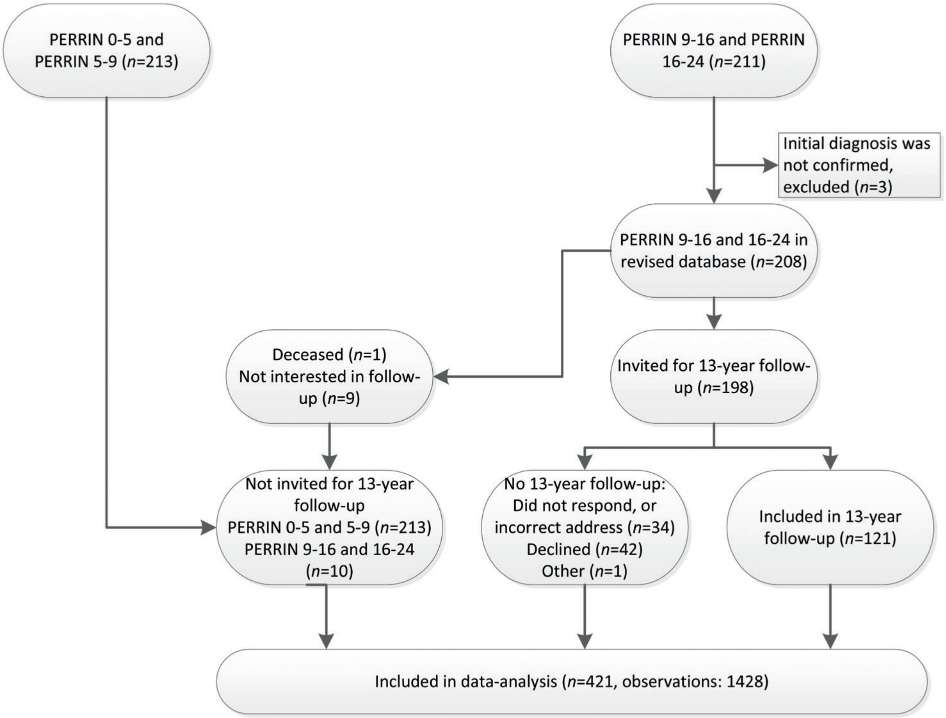
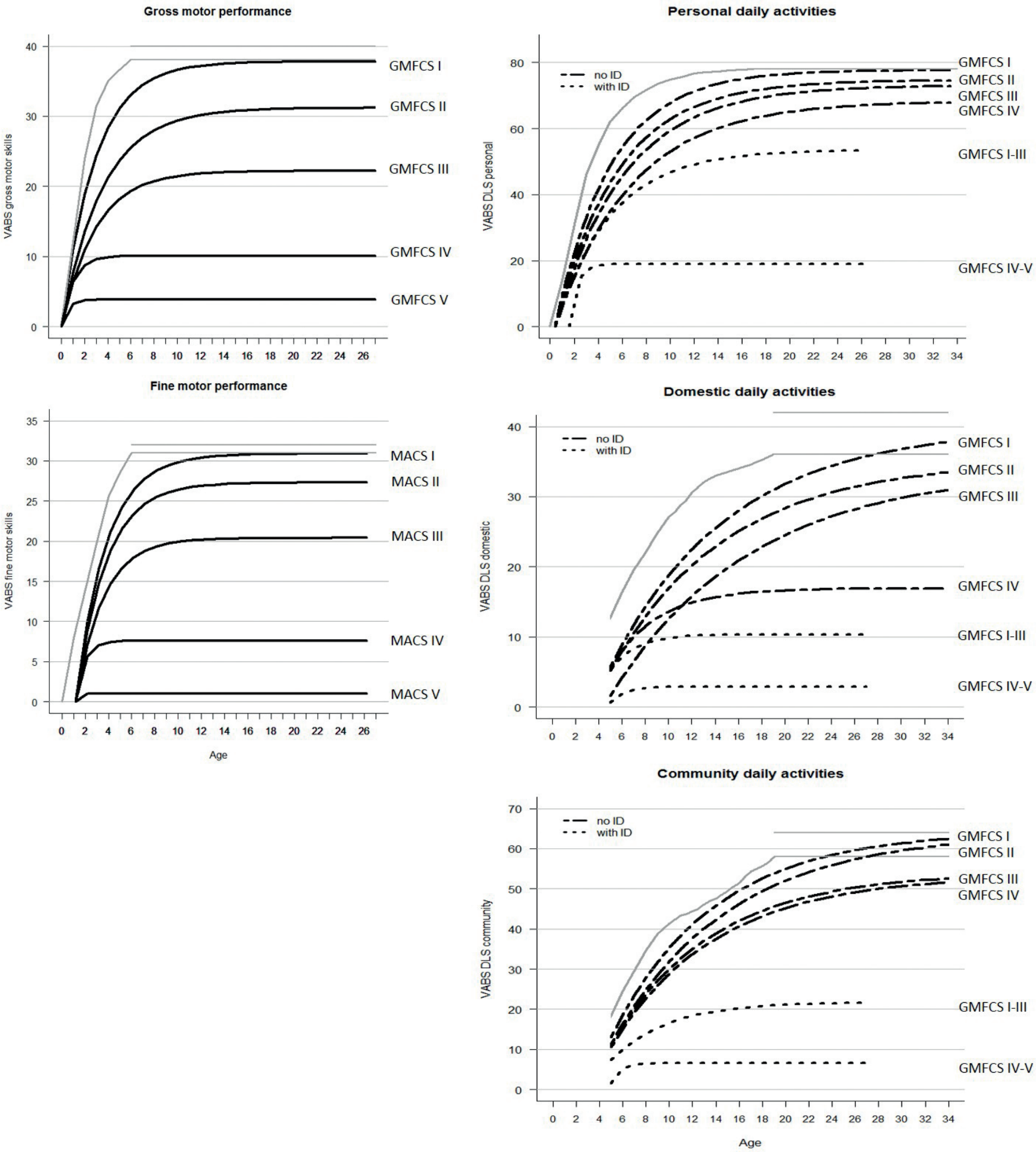


Figure 1: Flow diagram of inclusion of participants and observations.

Motor performance

The estimated limits of gross and fine motor performance showed a significant difference between all GMFCS and MACS levels, as the 95% CIs did not overlap. Limits were lower for each lower level of gross motor function or manual ability (Table 2).

The age_{90}s showed no significant difference between GMFCS and MACS levels I-III, and were estimated at 6-8 years (Table 2). Individuals with GMFCS and MACS level IV and V had a lower age_{90} compared to levels I-III, indicating that they reach their lower limit at a younger age compared with their less affected peers. Furthermore, both the large 95% CI's around the age_{90} and the raw observations of individuals with MACS level IV and GMFCS level V suggested that average development did not increase or decrease over time (Appendix 2).



Gross motor performance

Fine motor performance

Personal daily activities

Domestic daily activities

Community daily activities

Figure 2: Average curves of motor and daily activity performance development, with the age equivalents of the reference group indicated in gray.

Table 2: Model parameters of gross and fine motor performance development (1-27 years) for GMFCS and MACS level and parameters of performance of daily living skills development (1-34 years) for GMFCS level and intellectual status.

		GMFCS level	N	Mean no. of observations	limit	(95% CI)	50% range	Age_{90}	(95% CI)	Residual SDs	offset
Gross motor performance		I	134	3.4	37.8	(36.9-38.7)	35.8-39.8	6y8m	(6y1m-7y3m)	3.0	0.0
		II	44	3.3	31.2	(28.9-33.6)	28.5-33.9	8y1m	(6y6m-10y0m)	3.3	0.0
		III	53	3.2	22.2	(19.7-24.6)	18.9-25.4	6y8m	(5y0m-9y0m)	3.6	0.0
		IV	43	3.2	10.1	(8.8-11.4)	7.6-12.6	2y4m	(1y3m-4y3m)	2.2	0.0
		V	40	3.7	3.9	(3.3-4.4)	2.8-4.9	1y3m	(0y2m-9y7m)	1.5	0.0
		MACS level	**N**	**Mean no. of observations**	**limit**	**(95% CI)**	**50% range**	**Age_{90}**	**(95% CI)**	**Residual SDs**	**offset**
Fine motor performance		I	106	3.5	30.9	(29.7-32.2)	27.9-32.0	7y2m	(6y6m-7y11m)	2.2	1.2
		II	109	3.3	27.3	(25.6-29.0)	23.2-31.5	7y1m	(6y3m-8y0m)	2.5	1.2
		III	39	3.4	20.4	(17.0-23.8)	15.0-25.8	6y7m	(4y9m-9y3m)	2.7	1.2
		IV	24	3.3	7.6	(5.5-9.8)	4.7-10.5	2y11m	(1y5m-15y5m)	4.1	1.2
		V	23	3.4	1.0	(0.4-1.5)	0.2-1.7	1y6m	(1y3m-4y4m)	1.0	1.2
		GMFCS level	**N**	**Mean no. of observations**	**limit**	**(95% CI)**	**50% range**	**Age_{90}**	**(95% CI)**	**Residual SDs**	**offset**
Personal activity performance	no intellectual disability	I	194	3.3	77.8	(76.8-78.7)	75.2-78.0	11y2m	(10y9m-11y7m)	3.9	0.4
		II	42	3.2	74.7	(70.0-79.4)	68.5-78.0	12y5m	(10y11m-14y2m)	4.0	0.4
		III	36	3.3	73.1	(67.1-79.1)	66.0-78.0	13y8m	(11y10m-15y9m)	4.4	0.4
		IV	27	3.0	68.3	(61.7-74.8)	61.5-75.1	15y2m	(12y3m-18y9m)	6.0	0.4
	intellectual disability	I-III	44	3.0	53.6	(45.7-61.5)	45.6-61.6	11y2m	(8y6m-14y8m)	5.8	0.4
		IV-V	63	3.3	19.0	(15.7-22.3)	10.9-27.1	3y0m	(2y4m-4y5m)	6.8	1.6*

Table 2: Continued

		GMFCS level	N	Mean no. of observations	limit	(95% CI)	50% range	Age_{90}	(95% CI)	Residual SDs	offset
Domestic activity performance	no intellectual disability	I	168	3.3	39.9	(37.6-42.3)	36.1-42.0	27y5m	(24y11m-30y2m)	4.4	3.4
		II	34	3.3	35.2	(30.4-39.9)	32.6-37.8	26y8m	(21y10m-32y9m)	3.9	3.4
		III	26	3.4	34.0	(24.0-43.9)	28.6-39.3	32y5m	(22y6m-47y8m)	5.0	4.4*
		IV	21	3.0	16.9	(12.6-21.3)	12.1-21.7	12y9m	(7y5m-25y0m)	5.1	3.4
	intellectual disability	I-III	27	2.9	10.3	(7.9-12.8)	8.2-12.5	8y5m	(5y7m-14y10m)	3.5	3.4
		IV-V	40	3.7	2.9	(1.7-4.2)	0.6-5.3	6y6m	(4y9m-72y2m)	3.5	4.7*
		GMFCS level	**N**	**Mean no. of observations**	**limit**	**(95% CI)**	**50% range**	Age_{90}	**(95% CI)**	**Residual SDs**	**offset**
Community activity performance	no intellectual disability	I	168	3.4	64.4	(62.5-66.4)	60.8-64.0	23y4m	(22y2m-24y7m)	4.4	3.0
		II	34	3.3	64.0	(58.6-69.5)	60.8-64.0	26y5m	(23y3m-30y1m)	4.3	3.0
		III	26	3.5	54.1	(45.9-62.3)	46.6-61.6	22y11m	(18y10m-28y1m)	5.1	3.0
		IV	22	3.3	53.3	(45.6-61.0)	46.8-59.9	23y10m	(18y9m-30y5m)	6.0	3.0
	intellectual disability	I-III	27	2.9	21.8	(15.3-28.3)	15.1-28.5	14y3m	(9y10m-21y7m)	5.1	3.0
		IV-V	40	3.7	6.6	(3.9-9.4)	1.3-11.9	6y6m	(4y10m-90y8m)	4.3	4.8*

* If the offset, (determined based on all observations of a subdomain), resulted in a severity subgroup model that did not fit, the offset was based on the optimal fit to the observations of the subgroup

Daily activity performance

The limits of personal and domestic daily activity performance showed no significant difference between individuals with GMFCS levels I-III (without ID), while the limits for those with GMFCS level IV were lower compared to GMFCS level I (and II and III for domestic activity performance). The community daily activity limits of individuals with GMFCS level III and IV were lower than those with GMFCS level I. Inter-individual variance of daily activity performance limits (indicated by the 50% -ranges) tended to increase with increasing GMFCS level (Table 2).

The age_{90}s of individuals without ID (GMFCS level I-IV) showed no significant difference between GMFCS levels, except for individuals with GMFCS level IV who had a lower age_{90} for the community domain. The age_{90} ranged from 11-15 years for personal daily activities, from 26-32 years for domestic daily activities, and from 22-26 years for community daily activities.

In all domains of daily activity, the limits of individuals with ID were significantly lower than limits of individuals without ID. Among individuals with ID, the limits of those with GMFCS level IV-V were lower than those of GMFCS level I-III (Table 2).

Individuals with ID reached their limits earlier than individuals without ID, as was indicated by lower age_{90}s. This was significantly so for personal activities of individuals with GMFCS level IV-V and ID and for domestic and community activities of individuals with GMFCS level I-III and ID. Furthermore, for domestic and community activities, both the large 95% CI's around the age_{90}s and the raw observations of individuals with GMFCS level IV-V and ID suggested that average development did not increase or decrease over time (Appendix 2) .

DISCUSSION

This study describes the development curves of motor and daily activity performance from childhood into adulthood of individuals with CP by their functional ability level, to supplement the widely used development curves of motor capacity of children with CP.[8] Individuals with CP with walking ability (GMFCS levels I-III) approached their maximal performance levels at 6-8 years for gross and fine motor performance, at 11-14 years for personal (self-care) activities, at 26-32 years for domestic activities and at 22-26 years for community daily activities. Severely affected individuals showed the least favourable development of motor performance and daily activities, which was observed as lower maximal performance levels in all domains approached at a younger age.

The average maximal gross motor performance levels (limits) of children with CP were distinctly lower with each lower GMFCS level. While we observed a pattern for motor performance that is comparable to the published gross motor capacity curves, children with CP continue to improve their gross motor performance level as they age (age_{90} for GMFCS level I-III at 6-8 years), while their gross motor capacity has plateaud at a younger age (age_{90} at 4-5 years).[9] A similar effect of functional classification on the limits and similar slower development are observed when comparing the fine motor performance curves with those of fine motor capacity in a Swedish cohort.[10] However, direct comparison between these curves of performance and capacity requires caution, since the specific activities assessed in the outcome measures (GMFM or AHA for capacity and VABS for performance) are not identical.[17,26] Nevertheless, a lagged development of motor performance seems valid, as performance does not only depend on the ability of the person to do activities in a standardized setting (i.e. capacity), but also is affected by personal factors (such as motivation or self-efficacy) and the environment in which the activities are performed in daily life.[8,12,14] Therefore, health-care professionals should be aware of further development of motor performance in children with CP after they have reached their limit in motor capacity.

Development of self-care activities continued into adolescence, while domestic activities and community activities continued to develop into the early and late twenties. For these specific domains of activities the estimated maximal levels of performance of individuals with GMFCS level I-III without ID are close to those of typically developing individuals,[17] which is similar to our previous report on the overall daily activities domain.[15] Our results further confirm that development also occurs differently for specific domains of activities for individuals with CP. Regarding the pace of development, reference values of the VABS show that typically developing individuals reach 90% of their maximal performance level of self-care, domestic and community activity at about 7, 18 and 15 years of age, respectively.[17] This indicates that even though the limits are similar, the development of daily activity performance of individuals with CP without ID seems to be delayed, which is most pronounced in domestic activities.[17]

The ages up to which individuals with CP without ID were found to develop daily activities are surprisingly high. The ongoing development of daily activities well into adulthood supports the need for taking a life course health development approach for individuals with CP.[7,16] In addition to the lagged development of daily activity performance presented here, high proportions of this sample of individuals with CP experience difficulty in participation in domestic and community life in young adulthood.[27] Moreover, European adolescents with CP participate less frequently in domestic activities (doing chores) and community life (having work experience).[28] These aspects of difficulty and frequency of performance are also important to consider in clinical decision-making. Routine monitoring of individuals with CP for activity limitations or participation restrictions is recommended into adulthood, as development of performance of daily activities does not stop at age 18, when many individuals leave the pediatric setting and make the transfer to adult healthcare services.

The less favourable development of performance of those individuals with CP with significant impairments and individuals with CP and ID support findings of previous cross-sectional studies of younger populations.[4,13-15,29] This knowledge can be used to educate and counsel these individuals and their families on expected future daily activity performance. In addition, clinicians should take the development curves into consideration when setting treatment goals. For example, by focusing treatment on increasing their independence in daily activities or by promoting participation by creating opportunities while offering adequate supports and resources.

To further specify expectations of performance development and to identify subgroups at increased risk of less favourable development of activity performance, future studies should determine which personal factors besides CP-related factors such as GMFCS level, MACS level and ID and which environmental factors are determinants of performance development. In addition, it is of interest to examine which interventions and at what point during the development curve can best facilitate performance. For instance, we could evaluate the effects of a 'boost' of therapy with high intensity at an age when change in performance is expected, or the effects of offering participation-based therapy[30] when the development of activities stabilizes.

Limitations

A general limitation relates to the relatively small subgroups of those other than GMFCS level I without ID; there were in particular small numbers of observations for individuals with GMFCS levels II-IV, in the age of mid-twenties and up. Second, comparison with the VABS reference values is hampered by a different culture (USA) and timeframe (1984).[17] Furthermore, both caregiver-reported scores (for children and adults with ID) and self-reported scores (for adolescents and adults without ID) were used. However, we assume that the effect on the results will be limited, because of the straightforward content of the VABS items.

CONCLUSION

Development of motor performance of individuals with CP continues after gross motor capacity limits have been reached in childhood. Self-care performance continues into adolescence, and the development of domestic and community activities progresses into the mid-late twenties. Individuals with severe CP show the least favourable development of motor and daily activity performance.

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4

Development curves of communication and social interactions in cerebral palsy

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ABSTRACT

Aims: To determine development curves of communication and social interactions from childhood into adulthood for individuals with CP.

Method: This PERRIN-DECADE Study longitudinally assessed individuals with CP aged 1-20 years at baseline after 13-years (follow-up; n=421). Communication and social interactions were assessed using the Vineland Adaptive Behavior Scales. We estimated average maximum performance level (limit) and age at which 90% of the limit was reached (age_{90}) using nonlinear mixed-effects modeling.

Results: Individuals without intellectual disability (ID) were 21-34 years at follow-up. Limits of these individuals, regardless of GMFCS level, approached the maximum score and were significantly higher than those of individuals with ID. Age_{90}s ranged between 3-4 years of age for receptive communication, 6-7 years for expressive communication and inter-relationships, 12-16 years for written communication, 13-16 years for play and leisure and 14-16 years for coping. Individuals with ID were 21-27 years at follow-up. Individuals with ID with GMFCS-level V showed the least favourable development, but variation between individuals with ID was large.

Interpretation: Individuals with CP and without ID develop communication and social interactions similar to typically developing individuals, regardless of their level of motor function. Those with ID reach lower performance levels and vary largely in individual development.

INTRODUCTION

Over the past decades, the life expectancy of children with CP has substantially increased and is currently nearly comparable to the general population.[1] Nowadays, clinical and research interest focuses on understanding activity and participation outcomes. Children socially interact with their family, peers and others which contributes to their development toward optimal participation in adult roles. Receptive and expressive communication are considered essential for these social interactions; moreover, communication difficulties are associated with problems in social interactions with familiar partners.[2 3]

The Pediatric Rehabilitation Research in the Netherlands (PERRIN) program was set up to study the longitudinal development of activities and participation of individuals with CP.[4-7] Development of communication was described by type of CP and social interactions by GMFCS-level in a Dutch population of 421 individuals with CP towards young adulthood (1-24 years).[8 9] Individuals without ID eventually reached similar levels of communication and social interactions as typically developing individuals. Development of individuals with ID was studied up to age 16 and was found less favorable than that of individuals without ID. Communication curves were less favorable for individuals with Bilateral Spastic CP (BSCP) or Non-Spastic CP (NSCP) compared to individuals with Unilateral Spastic CP (USCP).[9] Social interaction curves were less favorable for individuals with GMFCS-level V compared to those with GMFCS-level I-IV.[8]

To better inform young individuals with CP and their families regarding their future functioning, the results of the PERRIN study may be improved in some aspects. In line with other studies that relate communication to GMFCS-level[10 11], the long-term development of communication may also be studied by GMFCS-level. In addition, our knowledge may gain clinical relevance by addressing aspects of social interactions in more detail. Furthermore, it is yet unknown at what age the maximal performance of communication and social interactions is reached. Motor development curves are widely used to monitor and predict the future gross motor capacity of individuals with CP.[12 13] These gross motor curves were established using a nonlinear mixed-effects model assuming a rapid development at first that slows toward reaching a (stable) limit. This model has also been used to estimate development curves for gross and fine motor- and daily activity performance and mobility and self-care capability in individuals with CP.[14 15] Using this model in the present study allows us to compare the long-term development in communication and social interactions with those of other outcomes.

Following up part of the PERRIN population 13 years after the first measurement provides the unique opportunity to 1) more accurately estimate the maximal performance and rate of development of communication and social interactions and 2) provide insight in the development of communication and social interactions in individuals without ID beyond the age of 24 years and in individuals with ID beyond the age of 16 years. Thus, the present study aims to determine the development curves of communication and social interactions into adulthood, in a Dutch population of individuals with CP aged 1-34 years. Based on the previous results for a smaller age range and the recent publication on other domains, we hypothesize that the development curves of individuals with CP without ID will reach similar limits compared to references, but that development is delayed.[9 14 16] Furthermore, we hypothesize that limits of development of individuals with ID will be less favourable compared to those of individuals without ID.

METHOD

This study was performed as part of the Dutch PERRIN-DECADE study, for which the participants of the two oldest cohorts of the PERRIN program have been followed up to 13 years after their last measurement. The recruitment process of the PERRIN program has been described in detail elsewhere.[4-7] In short, the program longitudinally measured 421 participants over 3 or 4 years between 2000 and 2007 in four age cohorts: PERRIN 0-5, PERRIN 5-9, PERRIN 9-16 and PERRIN 16-24. Individuals were invited when they had a confirmed diagnosis of CP. Individuals were excluded when they were diagnosed with additional disorders affecting motor functioning or when they or their caregiver lacked the basic knowledge of the Dutch language. In the PERRIN 16-24 cohort, individuals with ID (IQ<70) were also excluded.[4] Informed consent was obtained from each participant (or their parent or caregiver, in the case of participants with ID) and ethical approval from the medical ethics committees of each center.

A 13-year follow-up assessment of the PERRIN 9-16 and PERRIN 16-24 cohorts was completed in 2016, extending the database with observations of individuals without ID up to 34 years and individuals with ID up to 27 years. Figure 1 shows a flow-diagram of inclusion of observations in the data-analyses and Appendix 1 the number of observations by cohort. A trained researcher conducted face-to-face self-reported semi-structured interviews using the Dutch language version of the Vineland Adaptive Behavior Scales (VABS) survey.[17] The interviews took about 30 minutes per individual measurement. For the follow-up assessment, self-reported scores for individuals without ID and caregiver-reported scores were used for individuals with ID. The caregiver concerned the person who was most closely involved in the daily care of the individual with ID.

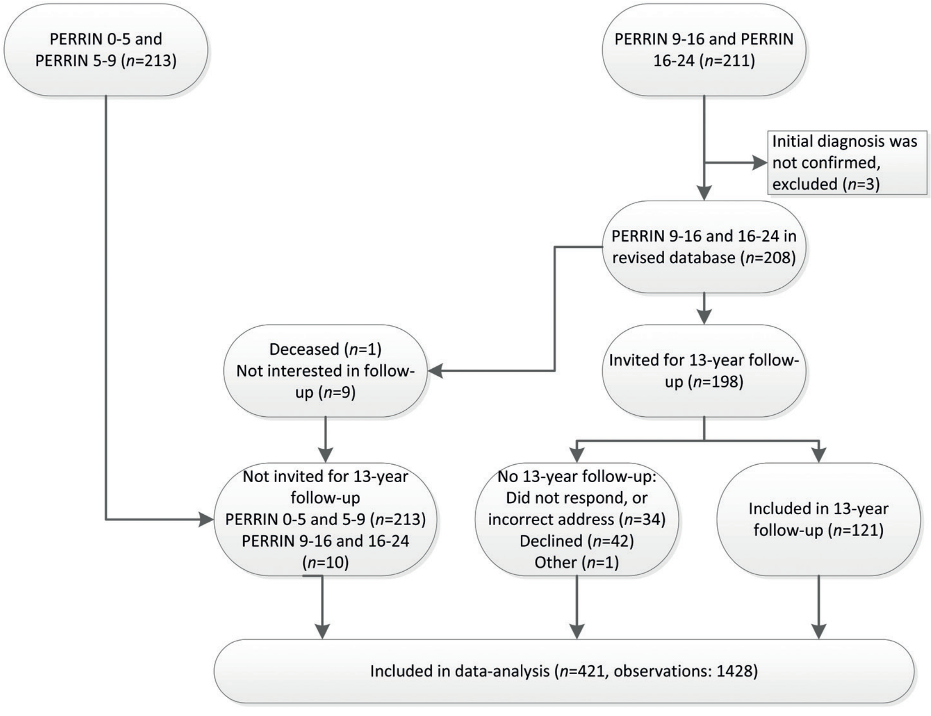

Figure 1: Flow diagram of inclusion

The VABS measure is a reliable and valid instrument to assess adaptive behavior of typical development between ages 0-19 years, suitable for individuals with and without disabilities.[17 18] The survey comprises 4 domains, among which communication and socialization. Activities are listed in developmental order with starting points for particular ages and have answer categories 0 (never performed) to 2 (usually or habitually performed). The domain of communication describes how an individual uses receptive communication (13 activities; score range 0-26), expressive communication (31; 0-62) and uses and understands written language (23; 0-26). The domain of socialization describes how an individual interacts with others (28 activities), plays or uses leisure time (20; 0-40) and demonstrates responsibility and sensitivity to others ('coping'; 18; 0-36). With respect to 'inter-relationships', 4 items regarding 'dating' were erroneously skipped in the 9-16 cohort and therefore discarded (score range 0-48). In agreement to the VABS guidelines, subdomain scores were considered as 'missing' if more than 4 items within the subdomain were missing.

The PERRIN 0-5 cohort completed the VABS screener covering relevant activities of the survey for this age group.[19] Inherent to their age, the screener did not include items on written communication. VABS screener scores were linearly transformed to reflect VABS survey scores. Reference values of typically developing individuals derived in north-west America[18] were compared to our results.

ID was defined as IQ<70 as assessed by the Snijders-Oomen Nonverbal Intelligence Test (PERRIN 0-5 cohort) or the Raven's Colored Progressive Matrices (PERRIN 5-9 cohort).[20 21] For the PERRIN 9-16 and 16-24 cohorts, individuals attending special education at a daycare center for children with severe disabilities or school for children with ID were classified with ID.[4 7] The level of gross motor function was assessed using the GMFCS, which addresses five categories of gross motor function: I (walks without limitations) to V (transported in a manual wheelchair).[22] Type of CP (USCP, BSCP or NSCP) was classified according to the Surveillance of Cerebral Palsy in Europe-guidelines.[23] Communication function was assessed using the CFCS, which addresses five categories of everyday communication performance: I (effective sending/receiving with unfamiliar and familiar partners) to V (seldom effective sending/receiving even with familiar partners).[24]

Descriptive statistics were performed in SPSS 22 (IBM SPSS Statistics, Armonk, NY, USA) and mixed-effects models were fitted using the 'nlme' package in R 3.2.5.[25] Appendix 2 presents details on the nonlinear model that we used. Development curves for each subdomain of communication and social interactions were determined separately for each GMFCS-level (communication, social interactions) and each type of CP (communication). Because of low number of observations, observations for individuals without ID with GMFCS-level V were discarded. With this asymptotic regression model, communication and social interaction subdomains were estimated as a function of age, approaching a limit of maximal potential performance with a rapid development rate at first but levelling off toward reaching the limit. The model has two parameters with straightforward clinical interpretations: the rate (speed of development) and limit (average maximal performance level). Random effects were included, accounting for dependency between repeated measurements within the same individual. To enhance interpretation, the rate parameter was transformed to age_{90}, i.e. the average age at which individuals reached 90% of their limit. The 95% confidence intervals (CIs) around age_{90} and limit were calculated; if the CIs around age_{90}s or limits of subgroups did not overlap, differences between these subgroups were statistically significant ($p<0.05$). From the random variance around the estimated limits for each subdomain of communication and social interactions, 50% ranges around the limits were constructed that cover the limits of 50% of the individuals in a subgroup. The residual SD of the models provide an indication of the model fit, with lower SD referring to a better fit. Since development does not start at birth for all subdomains, a start age of development was chosen based on the best model fit according to the Akaike Information Criterion, determined using all observations of the subdomain scores.[26]

RESULTS

Overall, 421 participants (73% without ID, 50% GMFCS-level I, 48% BSCP) contributed to 1,428 VABS observations of ages 1-34 years. At 13-year follow-up 121 adults with CP participated (21-34 years). Of those, 69% were classified as CFCS-level I. One individual without ID and four individuals with ID used a communication aid to communicate (i.e. communication device, communication cards / images and/or sign language). Table 1 presents the characteristics of the participants in each cohort. Figure 1 describes drop-outs which were not selective regarding sex or CP characteristics. Figure 2 shows the development curves by GMFCS-level. As the curves for individuals with GMFCS-levels I-IV were not significantly different and looked very similar, Figure 2 summarizes the four curves into one. Table 2 reports the corresponding limits, age$_{90}$ and 50% range around the limits. Finally, Appendix 3 presents raw observations, Appendix 4 parameters of development in communication by type of CP and Appendix 5 scatter plots by CFCS-level for individuals with ID.

In all subdomains, estimated limits were higher for individuals without ID compared to those with ID according to the 95% CIs (Table 2). For individuals without ID, estimated limits were comparable to that of typically developing individuals. The greatest lag compared to typically developing individuals was observed in written communication during childhood. The limits indicated that 9 year old children with CP scored 6 VABS points lower, meaning that they performed 3 VABS activities less than 9 year old typically developing individuals.

For individuals with ID, estimated limits for individuals with GMFCS-level I-IV were higher than for individuals with level V. This difference was significant for all subdomains, with the exception of written communication (Table 2), and may address the most difficult items, such as: giving complex directions to others, going to evening events with friends without adult supervision or independently weighing consequences of actions before making decisions.

For receptive communication, individuals with ID and GMFCS-level I-IV reach 90% and individuals with ID and GMFCS-level V 63% of the score of individuals without ID and GMFCS-level I. These proportions are 66% and 36% for expressive communication, 25% and 9% for written communication, 84% and 52% for inter-relationships, 68% and 38% for play & leisure and 53% and 22% for coping.

Compared to individuals without ID, inter-individual variances of the limits (50% ranges around the limits) were much wider for individuals with ID, for the communication subdomains in particular. With respect to social interactions, this was especially so for those with GMFCS-level V. These wide inter-individual variances indicate that the maximal development for individuals in this subgroup shows large variability.

The average age at which individuals with CP reached 90% of their limit in most cases did not differ significantly between individuals with and without ID nor between GMFCS-levels. For individuals without ID, the mean age$_{90}$ ranged between 3y3m-3y6m for receptive communication (versus 3y3m for typically developing individuals), between 6y1m-7y1m for expressive

Table 1: Participant characteristics of the study population and for subgroups at 13-year follow-up

	PERRIN 0-5	PERRIN 5-9	PERRIN 9-16		PERRIN 16-24	
	Baseline	**Baseline**	**Baseline**	**13y follow-up**	**Baseline**	**13y follow-up**
Number of participants	**97**	**116**	**107**	**67**	**101**	**54**
Number of observations	**314**	**328**	**399**	**67**	**260**	**54**
Age at baseline or 13-year follow-up, mean (SD)	1y7m (0y2m)	6y3m (1y0m)	11y2m (1y8m)	24y7m (1y7m)	18y6m (1y6m)	31y8m (1y5m)
Sex, *n* (%)						
Males	56 (58%)	76 (65%)	67 (63%)	45 (67%)	60 (59%)	29 (54%)
Females	41 (42%)	40 (35%)	40 (37%)	22 (33%)	41 (41%)	25 (46%)
Intellectual disability, *n* (%)						
No	50 (52%)	79 (68%)	75 (70%)	46 (69%)	101 (100%)	54 (100%)
Yes	45 (46%)	35 (30%)	32 (30%)	21 (31%)	0 (0%)	0 (0%)
	Unknown: 2	Unknown:2				
Level of gross motor function, *n* (%)						
GMFCS-level I	30 (31%)	56 (48%)	49 (46%)	30 (45%)	74 (73%)	38 (70%)
GMFCS-level II	13 (13%)	20 (17%)	14 (13%)	7 (10%)	8 (8%)	4 (7%)
GMFCS-level III	23 (24%)	17 (15%)	13 (12%)	8 (12%)	6 (6%)	4 (7%)
GMFCS-level IV	21 (22%)	9 (8%)	13 (12%)	9 (13%)	12 (12%)	7 (13%)
GMFCS-level V	10 (10%)	14 (12%)	18 (17%)	13 (19%)	1 (1%)	1 (2%)
Type of CP, *n* (%)						
USCP	41 (42%)	42 (36%)	37 (35%)	21 (31%)	41 (41%)	21 (39%)
BSCP	53 (55%)	56 (48%)	49 (46%)	31 (46%)	47 (47%)	25 (46%)
NSCP	3 (3%)	18 (16%)	22 (20%)	15 (23%)	13 (12%)	8 (15%)
Communication function, *n* (%)*						
CFCS-level I	*	*	*	37 (55%)	*	46 (85%)
CFCS-level II	-	-	-	17 (25%)	-	7 (13%)
CFCS-level III	-	-	-	9 (13%)	-	1 (2%)
CFCS-level IV	-	-	-	2 (3%)	-	0 (0%)
CFCS-level V	-	-	-	2 (3%)	-	0 (0%)

*CFCS was only available at 13y follow-up

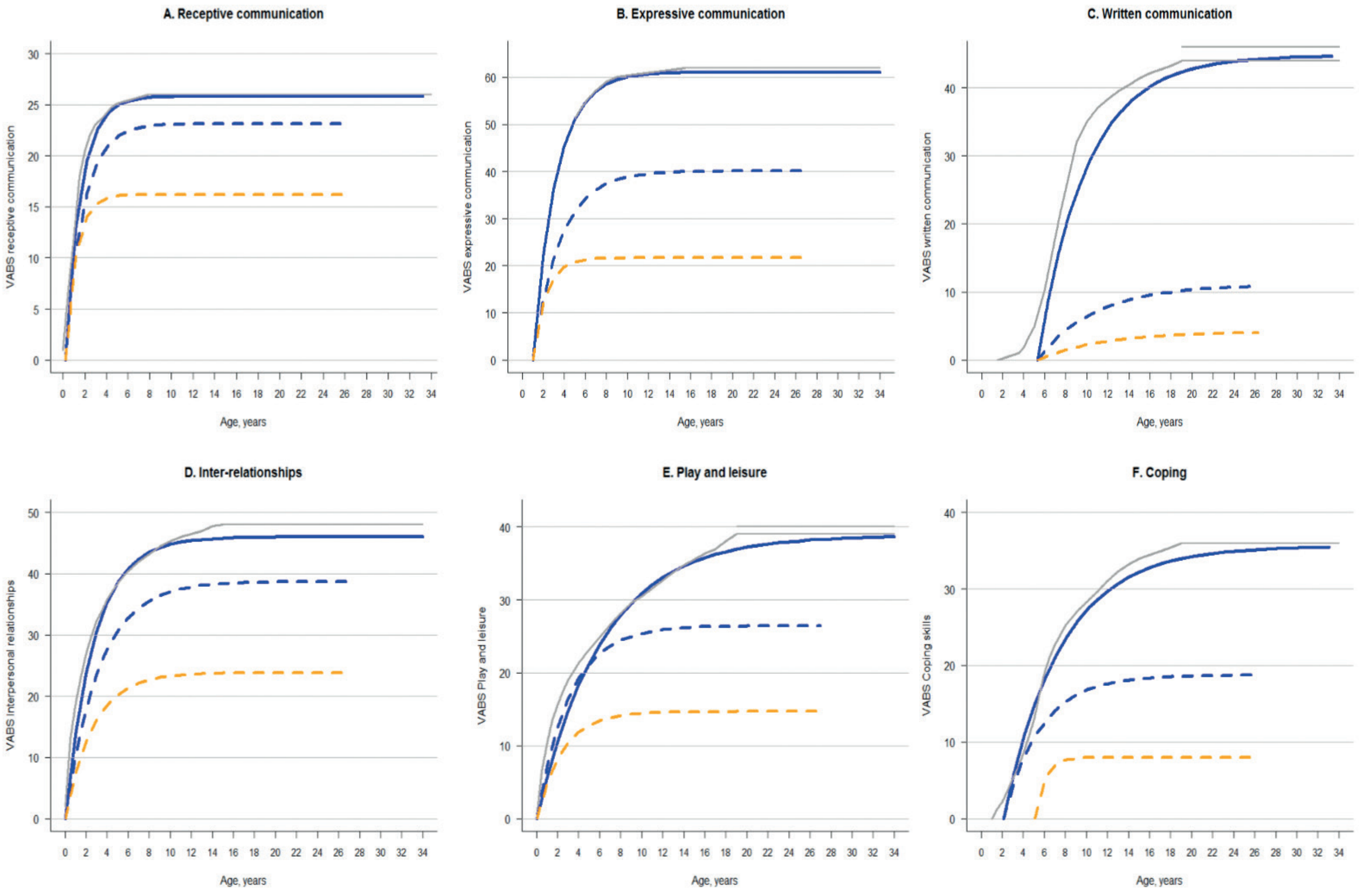

Figure 2: The development curves of communication and social interactions by ID and GMFCS-level

communication (versus 6y5m), between 12y9m-16y1m for written communication (versus 14y0m), between 6y3m-7y6m for inter-relationships (versus 7y10m), between 13y8m-16y7m for play and leisure (versus 15y0m) and between 14y0m-16y2m for coping (versus 13y0m).

Addressing maximal performance levels for receptive and expressive communication, individuals with ID and GMFCS-level I-IV reach their (lower) limits at a similar age as individuals without ID. Compared to individuals without ID and individuals with ID and GMFCS-level I-IV, individuals with ID and GMFCS-level V on average reach a lower limit at a young age, indicating poorer development. For written communication, the age_{90} of individuals with ID was higher than that of individuals without ID, but the large 95% CIs around the age_{90} (Table 2) and the raw observations (Appendix 3) indicate ample development for individuals with ID in this subdomain.

For play & leisure and coping, individuals with ID and level I-IV reach their lower limit on average at a younger age than individuals without ID, and individuals with ID and GMFCS-level V at an even younger age. However, large 95% CIs around the age_{90} of individuals with ID (especially those with GMFCS-level V) suggest that there was no rate of development to characterize average longitudinal curves.

DISCUSSION

In communication and social interactions, individuals without ID and GMFCS-levels I-IV follow development curves comparable to typically developing individuals. Development stabilizes in childhood for receptive communication, in adolescence for expressive communication and inter-relationships and in early adulthood for written communication, play & leisure and coping. GMFCS-level is only a marker of gross motor function. Although a poor gross motor function may affect communication and social interactions, it does not reflect communicative and social capabilities. Therefore, health-care professionals should not underestimate the communicative and social capabilities of young individuals with CP based on GMFCS levels. As expected, individuals with ID develop to lower maximal levels, and their development shows large individual variation.

The long-term follow-up of individuals with CP resulted in more accurate estimates of the maximal performance levels compared to previous PERRIN studies.[8 9] In addition, with the additional measurement, nonlinear mixed-effects models showed to fit the data well and enabled quantitative estimations of the rate of development and a more direct comparison with other development curves. Hence, not only the maximal level but also the rate of development of individuals with CP without ID seems comparable to reference data of typically developing individuals.[18] In interpreting this finding it should be noted that the study was carried out in the Netherlands, where the environmental context is relatively accommodating toward individuals with impairments and their families. Other studies have shown wide variations in social interactions across regions within European countries, ascribing an estimated one third of the unexplained variation in communication activities to variation between regions.[27] The finding indeed contrasts our hypothesis of delayed development and is different from the development of motor capacity, motor performance and daily activities in individuals with CP where limits are lower for those with lower levels of motor function and development seems delayed.[12 14] We conclude that individuals with CP without ID develop well in the domains of communication and social interactions, despite limitations in motor capacity and activity performance. This underlines the need to address the different activity and participation domains specifically.

Table 2: Parameters of development in communication and social interactions by ID and GMFCS-level

	GMFCS-level	*n*	limit	(95% CI)	50% range	Age$_{90}$	(95% CI)	Residual SDs	offset
Receptive communication									
no intellectual disability	I	194	25.8	(25.7-25.9)	25.8	3y6m	(3y4m-3y7m)	1.1	0.2
	II	42	25.6	(25.3-25.9)	25.1-26.0	3y6m	(3y3m-3y9m)	0.9	0.2
	III	36	25.7	(25.5-26.0)	25.7	3y3m	(3y0m-3y6m)	1.1	0.2
	IV	28	25.8	(25.5-26.0)	25.8	3y5m	(3y2m-3y9m)	1.1	0.2
intellectual disability	I-IV	72	23.1	(22.1-24.1)	21.0-25.3	4y0m	(3y6m-4y7m)	2.2	0.2
	V	37	16.2	(13.5-18.9)	11.0-21.5	2y6m	(1y6m-4y4m)	3.1	0.2
Expressive communication									
no intellectual disability	I	193	61.1	(60.6-61.6)	59.6-62.0	6y1m	(5y11m-6y3m)	2.2	1.0
	II	42	61.0	(59.5-62.4)	59.2-62.0	6y8m	(6y2m-7y2m)	3.7	1.0
	III	36	60.9	(59.3-58.7)	58.7-62.0	6y4m	(5y11m-6y10m)	2.7	1.0
	IV	28	61.1	(59.3-62.5)	58.7-62.0	7y1m	(6y7m-7y7m)	3.3	1.0
intellectual disability	I-IV	71	40.1	(34.6-45.7)	28.6-51.7	7y0m	(5y6m-9y0m)	5.7	1.0
	V	37	21.7	(14.5-28.9)	7.3-36.1	3y10m	(2y6m-6y6m)	5.3	1.0
Written communication									
no intellectual disability	I	166	44.7	(43.6-45.8)	42.3- 46.0	16y1m	(15y4m-16y10m)	4.4	5.3
	II	34	41.6	(37.5-45.6)	37.0-46.0	15y9m	(14y0m-17y11m)	4.0	5.3
	III	25	40.9	(35.0-46.7)	35.0-46.0	12y9m	(11y4m-14y6m)	4.3	5.3
	IV	20	37.2	(32.9-41.5)	31.7-42.7	15y0m	(13y0m-17y6m)	3.7	5.3
intellectual disability	I-IV	38	11.0	(6.3-15.8)	2.5-19.6	17y8m	(14y8m-21y8m)	2.9	5.3
	V	29	4.2	(0.7-7.6)	0-9.9	19y2m	(15y0m-25y2m)	2.5	5.3

Table 2: Continued

	GMFCS-level	*n*	limit	(95% CI)	50% range	Age$_{90}$	(95% CI)	Residual SDs	offset
Inter-relationships									
no intellectual disability	I	194	46.0	(45.4-46.6)	43.9-48.0	6y4m	(5y11m-6y9m)	2.4	0.0
	II	42	45.8	(44.4-47.2)	44.0-47.6	7y6m	(6y9m-8y4m)	2.8	0.0
	III	36	44.6	(43.0-46.2)	42.7-46.6	6y3m	(5y7m-7y1m)	2.9	0.0
	IV	28	46.1	(44.6-47.5)	44.1-48.0	6y10m	(6y1m-7y9m)	2.1	0.0
intellectual disability	I-IV	38	38.7	(36.0-41.4)	35.8-41.7	7y4m	(5y4m-10y3m)	4.1	0.0
	V	29	23.9	(18.7-29.0)	15.7-32.0	6y2m	(2y0m-19y2m)	4.7	0.0
Play and leisure									
no intellectual disability	I	194	38.8	(38.1-39.5)	37.1-40.0	14y6m	(13y10m-15y2m)	2.4	0.0
	II	42	40.3	(38.7-41.8)	39.5-40.0	16y7m	(15y2m-18y3m)	2.8	0.0
	III	36	37.7	(35.6-39.9)	35.8-39.7	13y8m	(12y1m-15y4m)	2.4	0.0
	IV	28	37.2	(35.1-39.4)	35.2-39.3	14y0m	(11y11m-16y5m)	3.1	0.0
intellectual disability	I-IV	38	26.5	(23.2-29.7)	23.2-29.7	10y4m	(7y2m-14y10m)	3.7	0.0
	V	29	14.7	(10.7-18.7)	8.4-21.1	5y8m	(1y0m-32y11m)	4.0	0.0
Coping									
no intellectual disability	I	192	35.6	(34.5-36.7)	32.7-38.4	14y8m	(13y9m-15y9m)	3.5	2.1
	II	42	36.6	(34.0-39.2)	34.3-38.9	16y2m	(14y3m-18y6m)	3.2	2.1
	III	36	34.4	(30.5-38.3)	30.9-37.9	15y7m	(13y0m-18y10m)	3.6	2.1
	IV	28	35.1	(32.6-37.5)	32.7-37.5	14y0m	(11y10m-16y8m)	3.6	2.1
intellectual disability	I-IV	38	18.8	(14.5-23.0)	13.6-23.9	10y2m	(6y5m-17y2m)	5.3	2.1
	V	29	8.0	(4.0-12.1)	0.9-15.1	7y1m	(5y2m-46y9m)	4.3	5.1*

* With respect to coping, the model for individuals with ID and GMFCS-level V did not fit when the offset was determined based on all observations. Therefore, the offset for this subgroup was based on the observations of the subgroup (5.1 years).

In line with previous PERRIN publications[8 9], development curves were less favorable for individuals with ID. The GMFCS-level was related to maximal performance levels of each subdomain. Those with GMFCS-level V performed poorer than those with GMFCS-level I-IV. As compared to other subdomains of communication, the average maximal performance level of receptive communication (e.g. following instructions requiring an action, listening to a teacher) was relatively high for individuals with ID. This suggests that individuals with ID perform relatively well in understanding spoken language. Possibly, their conversational partners adapt their message to an appropriate level. Also, a discrepancy may be present between understanding spoken language and what individuals are able to communicate as understood.[28] Lastly, individuals with ID and GMFCS-levels I-IV perform relatively well in inter-relationships (e.g. initiating conversations, buying gifts for someone). This finding may primarily reflect relationships with close relatives, as the VABS does not differentiate between relationships with familiar or unfamiliar people.

Inter-individual variability was much larger for individuals with ID, as indicated by larger 50% range around the limits and raw observations. This was particularly true for receptive and expressive communication, where some individuals with GMFCS-level V reached the maximum score, while others hardly developed and had very low scores. These differences are also reflected by broad distribution over CFCS-levels and MACS-levels, which stratification modes have earlier shown to strongly correlate with GMFCS-levels.[2 29] The large variation indeed seems partly attributed to communication function (see Appendix 5). In addition, it may be caused partly by different intellectual levels within those categorized as IQ<70. The large variation indicates a need for a personalized approach in rehabilitation for individuals with ID.

The present study has some limitations. The VABS lists activities in developmental order for children between 0-19 years. There may be relevant activities that develop after the ceiling-age. Therefore, development may be ongoing although a plateau on the VABS scores is reached. Since the most difficult items of inter-relationships were discarded, the activities in that domain reflect development between 0-15 years. Consequently performance may particularly develop after the reported limit is reached. Secondly, the VABS addresses *attendance* of participation by asking whether or not a person usually performs activities regardless the use of assistive devices, adaptations or supervision. Clinical experience and studies using concepts of participation other than *attendance*, such as *difficulty* of participation, or addressing other aspects, such as romantic relationships, indicate that individuals with CP do face restrictions in social interactions.[30 31] Furthermore, the present study discards the *quality* of communication and *satisfaction* or *preferences* in social interactions, which are relevant to consider in clinical decision-making. Thirdly, the VABS addresses objectively assessed basic skills. More complex skills or subjective aspects may develop differently. For example, individuals at risk for less favorable development of communication may additionally be assessed with more in depth examination of language, speech and motor function[32] and questioned on their subjective experience of social interactions. Finally, no Dutch reference data were available and comparison to reference data from the USA was descriptive rather than statistically tested.

Our findings may be used to inform individuals with CP on their future functioning in communication and social interactions. Health-care professionals may use the development curves for setting and adequate timing of specific treatment goals for individuals that seem to lag behind the expected development. Future studies should examine which factors measured in childhood predict communication and social interactions at adult age, especially for individuals with ID who currently show wide inter-individual variability.

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5

Long-term course of difficulty in participation of individuals with cerebral palsy aged 16-34 years: a prospective cohort study

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ABSTRACT

Aim: To determine the long-term course of difficulty in participation of individuals with cerebral palsy (CP) without intellectual disability (ID) between 16 and 34 years of age, described by Gross Motor Function Classification System level (GMFCS).

Method: 151 individuals with CP aged 16-20 years were included (63% male, GMFCS I-IV, without ID). The life habits questionnaire was used up to three times biyearly and at 13-year follow-up (13-year follow-up: *n*=98). Scores (range 0-10) reflect difficulty and assistance in participation in housing, education & employment, interpersonal relationships, recreation, community life, and responsibilities. Multilevel models were used to determine the course of difficulty in participation by GMFCS.

Results: Despite high average participation levels, 41 to 95% of adolescents and young adults with CP experienced difficulty. Difficulty in participation increased in most life areas in the mid- and late twenties, and for housing and interpersonal relationships from age 16 years onwards. In adolescents with GMFCS III and IV, difficulty in recreation and community life decreased up to age 23 years.

Interpretation: Individuals with CP experience increasing difficulties in participation in their mid- and late twenties. Clinicians should systematically check for participation difficulties in young adults with CP and timely offer personalized treatment.

INTRODUCTION

Cerebral palsy (CP) is a common childhood-onset physical disability causing activity limitation.[1] CP research and healthcare have traditionally focused on children, but as these children grow older, their participation in daily life changes. The transition from adolescence to adulthood is considered to be a critical phase to develop optimal participation in society.[2] Although healthcare services and education for children with CP are well developed, healthcare providers often struggle to offer appropriate care to support participation of this group in adulthood.[3]

Participation is defined by the International Classification of Functioning, Disability and Health (ICF) as ‘involvement in a life situation’ and encompasses, among others, the domains domestic life, education & employment, interpersonal interactions & relationships and community, social & civic life.[4] Studies on adolescents and adults with CP have demonstrated participation restrictions, i.e. problems individuals experience in involvement in life situations.[2,4-12] Some of these studies have addressed whether or not individuals actually performed certain life habits,[9-11] or how often they performed them in daily life.[7,12] According the ICF, participation performance can be qualified objectively by the experienced difficulty or the use of assistive devices or human assistance needed in performing life habits.[4] These qualifiers were used in cross-sectional studies showing that adolescents and young adults with CP may experience difficulty in participation, particularly in participation in education & employment and in community, social & civic life.[6,8,13,14] One study indicated that low proportions of adults with CP were dependent on human assistance in these participation domains.[14]

Although individuals with CP experience difficulty in participation during transition into adulthood,[2] no systematic knowledge is available on the long-term course of participation towards their late twenties or early thirties. At this age social roles are likely to change, from being dependent on ones parents to for instance living independently, being employed, and maintaining a stable relationship. These changes with increasing age are accompanied by growing social and environmental demands.[15] We therefore hypothesize that difficulty in participation increases for individuals with CP in their mid- or late twenties. The aim of the present study was to longitudinally investigate difficulty in participation in this age range by level of Gross Motor Function (GMFCS level), since level of function is known to be related to participation.[9,13,16] Furthermore, in the present study we focus on individuals *without* intellectual disability (ID), since participation trajectories differ greatly for individuals with ID.[9] These longitudinal curves may help individuals with CP and healthcare providers to timely focus on those aspects of participation where difficulties are experienced, in order to achieve optimal participation in adult life. Therefore, this study examines the long-term course of difficulty in participation of individuals with CP with GMFCS levels I-IV (without ID) aged 16-34 years.

METHOD

Participants

The long-term prospective cohort study PERRIN DECADE is part of the PERRIN (Pediatric Rehabilitation Research In the Netherlands) program. A 13-year follow-up assessment was performed of the PERRIN 9-16 cohort (baseline age: 9-13 years, *n*=108), with previous yearly assessments over the course of three years; and the PERRIN 16-24 cohort (baseline age: 16-20 years, *n*=103), with three previous biyearly assessments over the course of four years. The recruitment process for both cohorts is described elsewhere.[13,17] Eligible participants had a clinical diagnosis of CP and sufficient knowledge of the Dutch language to participate. In the PERRIN 16-24 cohort study, individuals with ID (roughly IQ<70) were excluded, since its primary focus was transition to an independent adult lifestyle.[13]

PERRIN DECADE recruitment

The study was approved by the medical ethical committees of VU University Medical Center and Erasmus MC University Medical Center. Appendix 1 presents a flowchart of the 13-year follow-up recruitment and observations included in the present study. Of all 211 participants of the PERRIN 9-16 and PERRIN 16-24 cohorts, 198 participants were invited for a 13-year follow-up assessment. The remaining 13 former participants were not invited because a) one had deceased, b) nine had indicated no interest in further participation, and c) three were excluded (their initial diagnosis could not be confirmed). Finally, 122 participants (62% response) gave informed consent and participated in the 13-year follow-up assessment. The follow-up data were added to the existing longitudinal PERRIN database, which was used for the present analyses.

The present study addresses difficulty in participation, for which data were collected only for individuals aged ≥ 16 years; thus, individuals without observations over age 16 were excluded (*n*=33). In addition, individuals with ID (*n*=21) or GMFCS level V (*n*=3) were excluded due to the small numbers of individuals in these categories with longitudinal observations. Thus, 151 individuals with CP (without ID) were included in the present analyses; of these, 105 (70%) were measured twice or more (see Appendix 2).

Measures

Life-Habits questionnaire

To assess difficulty in participation, an aspect of restricted participation, the Dutch version of the Life Habits questionnaire 3.0 general short form (Life-H) was administered. In addition to the domains addressing social roles (interpersonal relationships, education, employment, recreation, community life, and responsibilities), the housing domain was also assessed as we considered this to reflect a relevant domain of participation.[4,18] All domains contain 6-8 items, except for the education domain (3 items). The employment and education domains were combined into one score, since only one item of the education domain (either participation in high school or professional training) was applicable for each participant, and the employment domain includes two items that can reflect participation in education.

Items addressed two ICF qualifiers of participation performance: the difficulty experienced and the assistance needed with a life habit. Difficulty was recorded as ‘no difficulty’, ‘some difficulty’, ‘accomplished by a proxy’ or ‘not accomplished’. Assistance was recorded as ‘no assistance’, ‘use of assistive device’, ‘adaptation’ and/or ‘with human assistance’ (dependent functioning). If an item was applicable, a combined item score of 0-9 was assigned.[19] From item scores mean domain scores were calculated, which were linearly transformed into a 0-10 score, with 10 indicating optimal participation (see Appendix 3). Principal component analysis has shown acceptable unidimensionality within each domain,[20] thus supporting their use as an interval scale. Life-H domain scores ≥ 8.89 reflect independent functioning (i.e. functioning without human assistance) without difficulties, scores of 5.57-8.88 indicate independent functioning with difficulties, and scores ≤ 5.56 indicate dependent functioning or not able to accomplish (see Appendix 3).[21] Cut-off scores similar to earlier studies were used, after correcting for transformed score ranges.[13,20] The Life-H covers the ICF domains of participation well, is a valid instrument to measure performance of participation and has good inter- and intra-reliability.[19,22]

Characteristics of CP

Type of motor impairment was recorded as spastic (limb distribution: unilateral/bilateral), dyskinetic, ataxic, or mixed CP.[23] Level of motor functioning was recorded using the 5-level GMFCS, addressing five categories of gross motor function ranging from I (highest level) to V (lowest level); this has been validated for children and adults with CP.[24]

Statistical analysis

Descriptive statistics for baseline CP characteristics were performed using SPSS 22 (IBM SPSS Statistics, Armonk, NY, USA). Mean domain scores and proportions of participants experiencing difficulty (score < 8.89) and in need of human assistance (score ≤ 5.56) were described for observations in four age intervals: 16-18, 19-22, 23-26, and 27-34 years.

The longitudinal course of difficulty in participation for each Life-H domain was analysed using multilevel modelling in MLwiN 2.28 (Centre for Multilevel Modelling, Bristol University, UK), in which assessments were clustered within individuals. Age (continuous variable, range 16-34 years) and GMFCS level at baseline (categorical variable, GMFCS level I as reference category) were included as independent variables. Additionally, age^2 and the interactions of GMFCS level and age (and age^2, if applicable) were included in the model, if significant according to the Wald statistic. To account for individual variation, a random intercept and regression coefficient for age were included if they improved the model, evaluated by the likelihood ratio test. To check the analyses, models were repeated excluding individuals with only a single observation. To study possible decline of participation of individuals in their mid- and late twenties, additional analyses were done by repeating the multilevel models on the subgroup of observations across the age range of 23-34 years. A p-value <0.05 was considered significant for all analyses.

To study potential influence of dropouts the distribution of sex, GMFCS level and type of CP of individuals who dropped out (*n*=60) were compared to baseline (*n*=211) using a chi-square test.

RESULTS

Participant characteristics

The analyses included 379 observations of 151 individuals with CP without intellectual disability over the age range of 16-34 years. Most of the participants were male (63%), and most (71%) were classified in GMFCS level I (*n*=107), 11% in level II (*n*=16), 7% in level III (*n*=11) and 11% in level IV (*n*=17). The most common CP subtype was spastic CP (88%; 49% unilateral and 51% bilateral). Others had a dyskinetic (3%), ataxic (3%) or mixed type of CP (6%). Analysis showed that dropout was not selective regarding sex, GMFCS level, or type of CP. The additional analyses over age range 23-34 years were conducted on a subsample including 129 observations of 102 individuals. The distribution of gender, CP subtype and GMFCS level of this subsample was comparable to the full sample.

Descriptives of participation

Figure 1 presents the mean participation domain scores of the four age intervals for each GMFCS level (I-IV). Figure 2 presents the proportions of individuals experiencing difficulty or

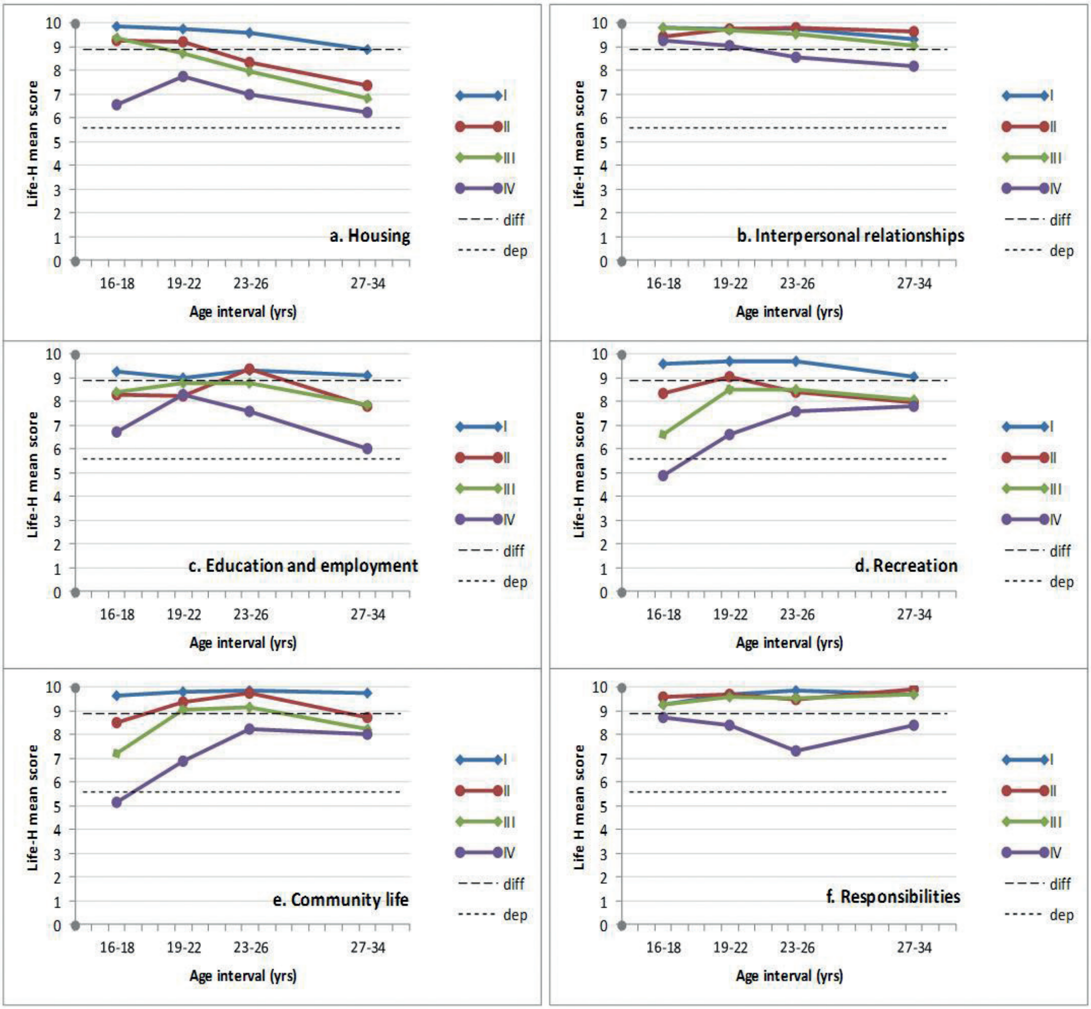

Figure 1a-f: Mean domain scores by GMFCS level and age intervals.
Number of observations at subsequent age intervals for GMFCS I: 73 (16-18 years), 112 (19-22 years), 49 (23-36 years), 41 (27-34 years); GMFCS II: 10, 14, 9, 4; GMFCS III: 8, 7, 6, 4; GMFCS IV: 11, 15, 8, 8. Dashed line (diff) represents criterion on independent functioning without difficulty (score ≥ 8.89), while dotted line (dep) at score 5.56 represents criterion of dependent functioning (score < 5.56).

needing human assistance in these age intervals by GMFCS level (for details see Appendix 4). Appendix 5 shows the proportions of individuals that considered each Life-H item applicable in the four age intervals; for most items, these proportions tended to increase with observations of increasing age intervals.

On average, individuals in GMFCS level I experienced no difficulty in participation in all domains (i.e. mean scores ≥ 8.89; Figure 1). Nevertheless, 41% of individuals in GMFCS level I, 77% in level II, 88% in level III, and 95% in level IV experienced difficulty in one or more of the participation domains. The highest proportions of difficulty were observed in individuals with GMFCS level IV, the oldest age range (23-34 years), and in the housing, education & employment and recreation domains (Figure 2).

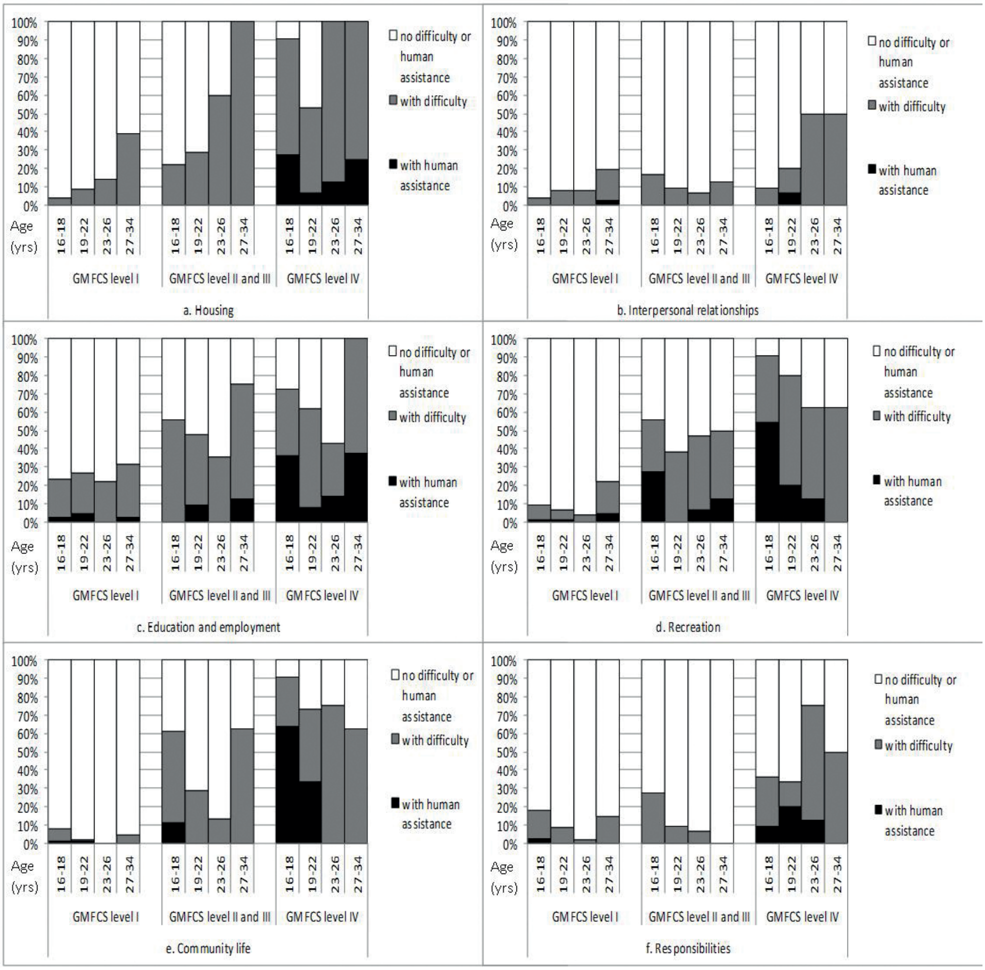

Figure 2a-f: Proportions of individuals with difficulty or needing human assistance by GMFCS level and age intervals.
Number of observations at subsequent age intervals for GMFCS I: 73 (16-18 years), 112 (19-22 years), 49 (23-36 years), 41 (27-34 years); GMFCS II and III: 18, 21, 15, 8; GMFCS IV: 11, 15, 8, 8.

Regarding dependency, Figure 1 shows that, on average, individuals with CP functioned independently in all domains (mean scores ≥ 5.56), except for adolescents (16-18 years) in GMFCS level IV in recreation and community life. Apart from adolescents in GMFCS level IV, small proportions of individuals in level II and III reported dependency in the participation domains (Figure 2).

Long-term course of participation

Table 1 shows the regression coefficients of the longitudinal models of the 6 participation domains over the entire age range (16-34 years). Random intercepts were included in all models. Random regression coefficients for age were included in the models of interpersonal relationships, recreation and community life. Table 2 shows the results of the additional analyses on participation domains of those individuals aged 23-34 years. Random intercepts were included in these additional models of housing, education & employment, and responsibilities, and no random regression coefficients for age were included. A ceiling effect was present in the participation scores on all domains; however, residuals of the models were normally distributed, allowing linear modelling.

Regression coefficients for age (and age^2) showed a decrease of housing and interpersonal relationships scores over age range 16-34 years (Table 1), indicating an increase of difficulty in participation on these domains. Additionally, a decrease of recreation scores was observed over age range 23-34 (Table 2). Regression coefficients for GMFCS showed that individuals in level IV scored lower (i.e. more difficulty) than individuals in level I on all domains (Table 1), except for education & employment of individuals in their mid- and late twenties (Table 2). Individuals in GMFCS level II scored lower than those in level I for education & employment, and community life, and those in level III for recreation and community life (Table 1). The course of difficulty in each participation domain is described in more detail below.

Participation in housing.

Housing scores decreased with increasing age for all GMFCS levels ($p<0.001$;Table 1). This decrease was stronger for individuals in levels II and III compared to level I, indicated by the significant interaction of age*GMFCS level II ($p=0.007$) and III ($p=0.002$). The additional analysis confirmed a significant decrease in housing scores with increasing age in the mid- and late twenties ($p<0.001$; Table 2).

Participation in interpersonal relationships.

Interpersonal relationships scores showed a quadratic association with age over the 16-34 age range (Table 1), indicating a decrease in score irrespective of GMFCS level which was more pronounced towards the end of the age range. The additional analysis confirmed a decrease of interpersonal relationships scores in the mid- and late twenties (Table 2).

Participation in education and employment.

Education & employment scores showed no significant relationship with age in the 16-34 age range (Table 1). Additional analysis showed a trend towards decreasing scores for those in GMFCS level I with increasing age in the mid- and late twenties. Furthermore, significant interactions between age and GMFCS level indicate a stronger decrease of scores for level II ($p=0.012$) and IV ($p=0.005$) compared to level I in participants in their mid- and late twenties (Table 2).

Participation in recreation.

Recreation scores showed a different course for GMFCS levels III and IV compared to the relatively stable course of level I, indicated by the significant interactions of age and age^2 with GMFCS level (Table 1). Figure 1 shows an initial increase of scores for GMFCS levels III and IV for age range 16-23. Additional analysis showed a decrease of scores in the mid- and late twenties, irrespective of GMFCS level ($p=0.008$; Table 2).

Table 1: Model regression coefficients of longitudinal course of six participation domains of the Life-H by GMFCS level

	Housing		Interpersonal relationships		Education & employment		Recreation		Community life		Responsibilities	
Associated factors	Regression coefficient (95% CI)	*p*-value Wald	Regression coefficient (95% CI)	*p*-value Wald	Regression coefficient (95% CI)	*p*-value Wald	Regression coefficient (95% CI)	*p*-value Wald	Regression coefficient (95% CI)	*p*-value Wald	Regression coefficient (95% CI)	*p*-value Wald
age	**-0.08 (-0.10 to -0.05)**	**<0.001**	**0.14 (0.02 to 0.26)**	**0.026**	-0.02 (-0.05 to 0.01)	0.203	0.22 (-0.04 to 0.48)	0.097	0.15 (-0.07 to 0.37)	0.187	0.00 (-0.02 to 0.0)	0.975
age^2	na		**-0.004 (0.001)**	**0.001 (-0.007 to -0.002)**	na		-0.005 (0.003)	0.046 **(-0.011 to 0.000)**	-0.003 (0.002)	0.187 (-0.007 to 0.001)	na	
GMFCS I	reference category		reference category		reference category		reference category		reference category		reference category	
GMFCS II	0.94 (-0.38 to 2.25)	0.164	-0.03 (-0.34 to 0.28)	0.850	**-0.64 (-1.19 to -0.10)**	**0.021**	0.46 (-7.68 to 8.61)	0.913	**-9.09 (-15.80 to -2.39)**	**0.008**	0.09 (-8.28 to -0.10)	0.731
GMFCS III	1.26 (-0.34 to 2.86)	0.123	-0.07 (-0.45 to 0.30)	0.705	-0.62 (-1.28 to 0.05)	0.068	**-16.24 (-26.48 to -6.00)**	**0.002**	**-15.84 (-24.43 to -7.25)**	**<0.001**	-0.12 (-0.71 to 0.47)	0.695
GMFCS IV	**-3.41 (-4.65 to -2.17)**	**<0.001**	**-0.72 (-1.02 to -0.42)**	**<0.001**	**-1.89 (-2.43 to -1.36)**	**<0.001**	**-14.83 (-23.48 to -6.18)**	**0.001**	**-16.25 (-23.91 to -8.60)**	**<0.001**	**-1.50 (-1.98 to -1.01)**	**<0.001**
age* GMFCS I	reference category		na		na		reference category		reference category		na	
age* GMFCS II	**-0.08 (-0.14 to -0.02)**	**0.007**	na		na		-0.10 (-0.80 to 0.60)	0.780	0.74 (0.17 to 1.31)	0.011	na	
age* GMFCS III	**-0.11 (-0.18 to -0.04)**	**0.002**	na		na		**1.13 (0.29 to 1.97)**	**0.009**	**1.15 (0.45 to 1.85)**	**0.001**	na	

Table 1: Continued

	Housing		Interpersonal relationships		Education & employment		Recreation		Community life		Responsibilities	
Associated factors	Regression coefficient (95% CI)	*p*-value Wald	Regression coefficient (95% CI)	*p*-value Wald	Regression coefficient (95% CI)	*p*-value Wald	Regression coefficient (95% CI)	*p*-value Wald	Regression coefficient (95% CI)	*p*-value Wald	Regression coefficient (95% CI)	*p*-value Wald
age* GMFCS IV	0.04 (-0.01 to 0.09)	0.147	na		na		**0.82 (0.11 to 1.54)**	**0.024**	**0.98 (0.35 to 1.61)**	**0.002**	na	
age^2* GMFCS I	na		na		na		reference category		reference category		na	
age^2* GMFCS II	na		na		na		0.002 (-0.013 to 0.016)	0.836	**-0.015 (-0.027 to -0.004)**	**0.011**	na	
age^2* GMFCS III	na		na		na		**-0.021 (-0.038 to -0.003)**	**0.018**	**-0.022 (-0.035 to -0.008)**	**0.002**	na	
age^2* GMFCS IV	na		na		na		-0.012 (-0.027 to 0.002)	0.093	**-0.016 (-0.029 to -0.004)**	**0.011**	na	

bold font: significant regression coefficient according to the Wald statistic (p<0.05)
na: variable did not meet criteria for inclusion (was not significant according to the Wald statistic (p<0.05)

Participation in community life.
Community life scores showed a different course for GMFCS level II-IV compared to level I (significant interactions of age and age^2 with GMFCS level; Table 1). An increase of scores for levels II-IV is visible for the age range 16-23 (Figure 1). Additional analysis decreasing scores in the mid- and late twenties of individuals with GMFCS level I with age and a significant interaction of age and GMFCS, indicating a stronger decrease in level II compared to level I (Table 2).

Participation in responsibilities.
For responsibilities, scores showed no significant relationship with age in the 16-34 age range. Additional analysis showed a different course (i.e. a larger increase) in the mid- and late twenties of GMFCS level IV compared to level I ($p<0.001$; Table 2).

DISCUSSION

This is the first study to present the long-term course of difficulty in participation of individuals with CP without ID from adolescence into their late twenties and early thirties, described by GMFCS level (I-IV). In their mid- and late twenties, these individuals showed increasing difficulty in 5 of the 6 participation domains. Individuals in GMFCS level III and IV showed an improvement in participation in recreation and community life during adolescence and early adulthood.

As hypothesized, difficulty in participation increased with increasing age in the mid- and late twenties in all domains (except for responsibilities); this may be due to an age-related change of desired or socially defined roles when developing into full adulthood,[15] such as expectations regarding living situation, employment, and maintaining relationships. This mechanism, when taking on adult roles or increasing expectations of the young adults and their environment lead to more difficulties, can be indicated as 'growing into a deficit'.[25] This assumption is supported by an increase in the proportion of the Life-H items considered relevant to participants with increasing age (Appendix 5). No longitudinal studies were available for comparison. Cross-sectional studies in adult populations, however, found no significant relationship between difficulty in participation and age.[5,14] Still, Boucher et al. also argued younger participants seemed to participate without difficulty more often.[5] The discrepancy with the present results might be attributed to the specific age window and/or larger sample size of the study, therefore focusing more specifically on the phase of development towards adult roles, when demands and expectations are assumed to change. Future research may investigate this assumption by studying the possible discrepancy between expectations for adult life of individuals with CP and their actual participation performance. For now, our results indicate that attention is required for participation difficulties of individuals with CP in their twenties.

Our results add to the evidence that individuals with CP who are more severely physically impaired experience more problems in participation compared to those with milder impairment.[5,9,10] Furthermore, in recreation and community life, individuals with lower levels of motor functioning continued to develop their level of participation into their early twenties, while those with higher levels of motor functioning reached maximal levels at younger age. As a consequence, individuals in GMFCS level IV reached independence in participation at older age. In contrast, these individuals have been found to reach their motor capacity limits younger than less affected peers, or even decline in motor capacity in their teens.[26] Furthermore, according to their level of motor functioning, they require human assistance in specific movements, e.g. in making transfers.[24]

Table 2: Additional models over 23 years of age: Model regression coefficients of longitudinal course of six participation domains by GMFCS level

	Housing		Interpersonal relationships		Education & employment		Recreation		Community life		Responsibilities	
Associated factors	Regression coefficient (95% CI)	*p*-value Wald	Regression coefficient (95% CI)	*p*-value Wald	Regression coefficient (95% CI)	*p*-value Wald	Regression coefficient (95% CI)	*p*-value Wald	Regression coefficient (95% CI)	*p*-value Wald	Regression coefficient (95% CI)	*p*-value Wald
age	**-0.11 (-0.15 to -0.07)**	**<0.001**	**-0.06 (-0.10 to 0.02)**	**0.003**	-0.05 (-0.11 to 0.01)	0.085	**-0.07 (-0.12 to -0.02)**	**0.008**	-0.03 (-0.055 to 0.003)	0.081	-0.02 (-0.05 to 0.01)	0.27
GFMCS I	reference category		reference category		reference category		reference category		reference category		reference category	
GMFCS II	**-1.25 (-1.83 to -0.68)**	**<0.001**	0.20 (-0.31 to 0.71)	0.437	**5.48 (0.91 to 10.05)**	**0.019**	**-1.16 (-1.83 to -0.50)**	**0.001**	**2.72 (0.28 to 5.17)**	**0.029**	-0.55 (-2.77 to 1.67)	0.627
GMFCS III	**-1.83 (-2.47 to -1.19)**	**<0.001**	-0.22 (-0.79 to 0.35)	0.556	-0.92 (-6.01 to 4.17)	0.723	**-1.08 (-1.82 to -0.33)**	**0.005**	-0.70 (-1.83 to -3.22)	0.588	-0.91 (-3.46 to 1.63)	0.482
GMFCS IV	**-2.58 (-3.11 to -2.06)**	**<0.001**	**-1.16 (-1.63 to -0.70)**	**<0.001**	3.46 (-0.84 to 7.75)	0.115	**-1.69 (-2.29 to -1.08)**	**<0.001**	**-2.17 (-4.19 to -0.15)**	**<0.001**	**-5.80 (-7.91 to -3.68)**	**<0.001**
age* GFMCS I	na		na		reference category		na		reference category		reference category	
age* GMFCS II	na		na		**-0.22 (-0.38 to -0.05)**	**0.012**	na		**-0.12 (-0.21 to -0.03)**	**0.012**	0.01 (-0.07 to 0.09)	0.757
age* GMFCS III	na		na		0.00 (-0.18 to 0.18)	0.997	na		-0.06 (-0.16 to 0.03)	0.171	0.03 (-0.07 to 0.12)	0.302
age* GMFCS IV	na		na		**-0.22 (-0.37 to -0.07)**	**0.005**	na		0.02 (-0.06 to 0.09)	0.646	**0.14 (0.06 to 0.22)**	**<0.001**

bold font: significant regression coefficient according to the Wald statistic ($p<0.05$)

na: variable did not meet criteria for inclusion (was not significant according to the Wald statistic ($p<0.05$)

The present results indicate that participation needs prerequisites other than mobility or motor capacity, and while level of GMFCS is an important determinant for participation, there are also other factors to consider. Assistive devices or adaptations, for instance, may successfully contribute to independent participation of individuals with GMFCS level IV.

We found high average levels of participation; however, substantial proportions of individuals with CP experienced difficulty and some needed human assistance in participation. The domains domestic life, education & employment and recreation deserve special attention, since the proportions of difficulty were particularly high in these domains.[6,14] The discrepancy between high average levels of participation versus large proportions of individuals with CP experiencing difficulty, indicates a need to systematically check for participation difficulties in order to offer appropriate personalized care.

Strengths and limitations

Due to the exclusion of individuals with ID and the dropouts during follow-up, this study included low numbers of participants in GMFCS level II-IV (especially in the oldest age interval) which increases uncertainty. The additional analyses over age-range 23-34 years lacked the added value of longitudinal research, since most observations in this age range were from the 13-year follow-up only and were, therefore, of a cross-sectional nature. Furthermore, the Life-H domain scores were averaged from 6 to 8 ordinal items. These scores were assumed to be at the interval level, which was previously supported by their unidimensionality.[20]

We are aware of an ongoing discussion regarding the concept of participation, with different approaches to define participation, including also more subjective aspects of participation (such as: involvement) and participation-related constructs (such as: activity competence).[27,28] In the present study we used the ICF definition of participation with its qualifiers, assessing performance objectively. Accordingly, the Life-H domain scores were designated as participation outcomes. Some of the items may be considered activities rather than participation (e.g. moving around within your home), but the majority of items were classified to assess participation.[29] Our results provide valuable insights for individuals with CP and rehabilitation clinicians regarding experienced difficulty in participation. Future studies may add to these regarding subjective aspects of participation, reflecting individuals' preferences or satisfaction with participation.

Clinical implications and recommendations

Clinicians should be aware that increasing proportions of individuals with CP in their twenties and early thirties may experience difficulties in participation, particularly in domestic life, education & employment, and recreation. These individuals may benefit from systematic screening of potential participation problems and adequate support or training in several life areas, e.g. supporting them towards independent living or when entering the labor market.[30] Our results address difficulty in participation performance. Other aspects of participation, such as frequency of participation or an individual's subjective experience of participation are also important to consider in clinical decision-making. Specific consideration is needed for individuals in GMFCS level IV in order to set realistic expectations for future functioning, since they continuously experienced more difficulties than their peers in adolescence and adulthood. Future studies might extend the present results by investigating the subgroup of individuals with CP *with* ID, since ID is an important factor limiting participation,[8,9] and by estimating the influence of personal and environmental factors on difficulty in participation.

CONCLUSION

Difficulty in participation increases in the mid- and late twenties of individuals with CP without ID. Although individuals with CP have high mean levels of participation, many still experience difficulty in participation. Thus, systematic screening and timely support seem necessary to improve the development of optimal participation of young adults with CP.

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6

Autonomy in participation of individuals with cerebral palsy from youth into adulthood

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ABSTRACT

Aim: To determine the long-term development of autonomy in participation of individuals with cerebral palsy (CP) without intellectual disability (ID).

Method: Individuals with CP were assessed cross-sectionally (46%) or up to four times (54%), between ages 12-34 years. Autonomy in participation was classified using phase 3 of the Rotterdam Transition Profile. A logistic GEE regression model was used analyzing autonomy in six domains (independent variables: age, GMFCS level and interaction age*GMFCS level). Proportions of autonomy were compared to references using binomial tests ($p<0.05$).

Results: In most domains, over 90% of participants (*n*=189, 400 observations, 80% GMFCS I-II) reached autonomy in participation in their late twenties, except for intimate/sexual relationships. Those with GMFCS III-V compared to I-II had less favorable development of autonomy in transportation, intimate relationships, employment and housing, and more favorable development in finances. Compared to references, fewer individuals with CP were autonomous in participation.

Interpretation: In most participation domains, 90% of individuals with CP without ID eventually achieve autonomy. Nevertheless, they are less often autonomous than peers, and individuals with GMFCS III-V demonstrate less favorable development. This knowledge of autonomy may guide expectations of youth with CP and their caregivers. Furthermore, rehabilitation professionals should address autonomy development in intimate relationships, employment and housing, especially in individuals with lower gross motor function.

INTRODUCTION

During their lifespan, individuals need to make several transitions that demand adaptability (e.g. entering secondary school, or having a first romantic relationship or job). From this perspective, emerging adulthood is a challenging period since young persons have to adapt to participating in new social roles and more complex life situations.[1] Cardol et al. highlighted the importance of autonomy in participation,[2] which can be described for several domains, such as domestic life, interpersonal interactions and relationships, major life areas (education, employment, and economic life), as well as community, social, and civic life.[3] Optimally, participation in these domains includes the freedom of decision-making and acting based on one's own attitudes and reasoning. These two latter constructs are also called decisional autonomy and executional autonomy.[2] Achieving independence and developing self-determination in participation in adult roles, might protect a person from disadvantaged participation outcomes and poor quality of life on the long term.[4]

Development towards adult roles may provide additional challenges for individuals with a childhood-onset disability, such as cerebral palsy (CP). Individuals with CP have disorders of movement and posture that are often accompanied by cognitive, behavioral or other impairments, caused by non-progressive brain disturbances.[5] Nowadays, since almost all children with CP reach adulthood,[6] the transition into adulthood has gained ever-increasing interest in pediatric care.[7] Although knowledge on the development of autonomy in participation of individuals with CP from youth into adulthood is limited, it could improve rehabilitation care as autonomy in participation is considered an important goal of rehabilitation care.[2]

In a previous phase of the PEdiatric Rehabilitation Research In the Netherlands (PERRIN) study, the Rotterdam Transition Profile (RTP) was found to validly assess an individual's phase of transition from being dependent on adults towards a self-reliant autonomous life, in six domains of participation.[4] Individuals with CP in their early twenties were found to lag behind in autonomy compared to typically developing peers.[4] Furthermore, individuals with a lower level of gross motor functioning were less often autonomous in participation compared to those with a higher level.[4,8] Knowledge on the development of autonomy in participation over time is limited, since only cross-sectional studies and one study with a two-year follow-up, are available for individuals with CP up to age 25 years.[7] Therefore, it remains unknown whether the development of autonomy over time differs between subgroups of individuals with CP, such as those with lower versus higher levels of gross motor functioning. Furthermore, regarding their autonomy, it is unknown whether individuals with CP continue to lag behind their age mates up to their early thirties.

Insight into the development of autonomy in participation of individuals with CP on the long term can guide expectations of youth with CP and their parents, and may identify targets for rehabilitation care. The present study focuses on individuals *without* intellectual disability, since they are expected to have the capacity to reach autonomy in adult life. For those with intellectual disability adult roles may differ greatly, presenting other developmental goals which may require a different approach.[4]

This study aimed to: i) describe the long-term development of autonomy in individuals with CP in several domains of participation from their teens into early thirties and examine differences in development between individuals with high/low levels of gross motor function; and ii) compare proportions of individuals with CP who are autonomous in participation to reference values of typically developing peers in the Netherlands.

METHOD

Study design

This study is part of the Dutch PERRIN program. Participants were longitudinally assessed between 2000 and 2007 in four age cohorts; PERRIN 0-5 (baseline age: 1-2 years), PERRIN 5-9 (baseline age: 5, and 7 years), PERRIN 9-16 (baseline age: 9, 11, and 13 years) and PERRIN 16-24 (baseline age: 16-20 years, 3 biennial assessments). The cohorts were considered representative of the population of individuals with CP, since all those known in rehabilitation care in the respective age ranges were invited to participate in the PERRIN study, and we assume that in the Netherlands almost all children with CP are known in rehabilitation care. Two long-term follow-up assessments were recently performed: PERRIN Participation in Perspective (PiP), a 10-year follow-up study of PERRIN 0-5 and 5-9, and PERRIN DECADE, a 13-year follow-up of PERRIN 9-16 and 16-24.[9,10] Observations from these follow-up studies and the previous biennial assessments of the PERRIN 16-24 cohort contributed to the present study; previous observations of PERRIN 0-5, 5-9 and 9-16 did not include assessments of autonomy in participation and consequently were added as cross-sectional data. Data were collected during interviews in the home situation, except in the PERRIN PiP follow-up study (participant age: 12-17 years) that collected data using online or paper-based surveys.

Participants

Details of the recruitment process of the four age cohorts are described elsewhere.[11,12,4,13] For the present study, eligible participants had a clinical diagnosis of CP without intellectual disability, no additional diagnosis affecting gross motor function, and they or a proxy were able to understand Dutch interviews or questionnaires. Individuals were classified with intellectual disability if they had an IQ below 70 assessed by the Snijders-Oomen Nonverbal Intelligence Test (PERRIN 0-5),[14] or Raven's Colored Progressive Matrices (PERRIN 5-9),[15] or based on educational level: those following a special education program for individuals with severe intellectual disability (PERRIN 9-16 and 16-24).[11] Former participants of the cohorts were recruited through information letters.[9,10] In total, 189 participants were included: 143 participated in the long-term follow-up studies, 46 PERRIN 16-24 participants who did not participate in the 13-year follow-up were included from the existing PERRIN database (Supporting Information (SI) 1). Of all participants, 90 were assessed longitudinally; either up to 4 years (35 individuals) or up to 13 years (55 individuals).

Ethical considerations

Approval for the cohort studies was obtained from the Medical Ethics committees of all participating centers. The Medical Ethics Committees of UMC Utrecht (for PERRIN PiP), Amsterdam UMC, location VUmc, Amsterdam and Erasmus MC Medical Center, Rotterdam (for PERRIN DECADE) agreed the long-term follow-up studies were outside the scope of the Medical Research Involving Human Subjects Act. All participants, or their legal representatives, provided informed consent.

Measures

Autonomy in participation

The development of autonomy in participation was monitored with the Dutch-language version of the RTP, which is validated to classify phases of transition into adulthood.[4] Decisional and executional autonomy are addressed in combination with the phases of transition for six domains of participation: leisure, transportation, finances, intimate/sexual relationships, education and employment (hereafter referred to as employment), and housing. For each domain, the most appropriate phase of transition was scored; either having no experience (phase 0), being dependent on adults (phase 1), experimenting and orientating to the future (phase 2), or being self-reliant or autonomous (phase 3). Phase 0 is not applicable for the domains housing and transportation.[1] Specific descriptions for the transition phases for each domain are shown in SI 2. Autonomy in participation (phase 3) specifically referred to: going out in the evening with peers (leisure), organizing transportation independently (transportation), being economically independent; paid job or receiving disability benefits (finances), having had experience with sexual intercourse (intimate relationships), having a job; sheltered, paid or unpaid employment (employment), and living independently; including assisted living while making one's own decisions regarding domestic life (housing). SI 2 presents the 2011 version of the RTP, of which the Dutch translation was used for the long-term follow-up.

Demographic and clinical characteristics

Age and level of education were recorded at each assessment (with exception of level of education for the PERRIN PiP cohort). Three levels of education were distinguished according to the International Standard Classification of Education 2011: low (lower secondary education or lower), medium (upper secondary education and post-secondary non-tertiary education), and high (short-cycle tertiary education, higher professional education, or university).[16] Type of motor impairment and level of gross motor function were recorded at baseline. Type of motor impairment was classified as spastic unilateral, spastic bilateral or mixed (dyskinetic, ataxic, and mixed CP).[17] The Gross Motor Function Classification System (GMFCS) was used to classify motor function in five levels ranging from walking without limitations (level I) to severe limitations in self-mobility (level V).[18]

Statistical analysis

Descriptive statistics were computed for demographic and clinical characteristics, and for the distribution of RTP transition phases at different ages (12-14, 15-18, 19-22, 23-26, 27-30, and 31-34 years). In addition, the age when 50% of the sample had reached autonomy in participation (phase 3) was described. For the remainder of analyses the transition phases were dichotomized (phase 0-2 versus phase 3 [autonomy]). The GMFCS levels were also dichotomized (I-II versus III-V).[19] A logistic generalized estimating equations (GEE) regression model was used for the outcome autonomy (yes/no) in each specific domain. Independent variables were age, GMFCS level (I-II as reference category), and the interaction between age and GMFCS level. The model adjusted for dependency of observations within one subject and also allowed cross-sectional assessments since it appropriately handles missing data. The model provided estimates of the effect of GMFCS level (I-II versus III-V), age, and the interaction of these on the odds of having achieved autonomy over time. For a correct interpretation of odds ratios (OR), age was standardized by subtracting the mean. An OR >1 indicates larger odds of autonomy, while an OR<1 indicates smaller odds of autonomy. The OR for GMFCS III-V indicates the difference in odds of those with GMFCS level III-V compared to those with GMFCS level I-II, for age the OR indicates the odds for each year an individual with GMFCS I-II is older, and finally the interaction indicates the odds of autonomy for each year and individual with GMFCS level III-V is older.

Autonomy in participation of individuals with CP was compared with that of Dutch age-matched reference data using two-tailed one-sample binomial tests in 5-year age intervals: 15-19, 20-24, 25-29, and 30-34 years. Data of the general population were extracted from the database of Statistics Netherlands (StatLine) for finances (income through employment or benefits), and housing (living independently);[20,21] reference data for intimate relationships (sexual intercourse within the last 12 months) were obtained from Rutgers/Soa Aids Nederland.[22] No suitable reference data were available for employment, leisure, and transportation. Analyses were performed using SPSS, version 24.0 (IBM SPSS Statistics, Armonk, NY, USA); level of significance was $p<0.05$.

RESULTS

The 189 participants contributed to 400 observations (SI 3). See number of observations by outcome (and age category) in Table I and II and Figure 2. Participants' age ranged from 12-34 years, 62% were male, 80% were GMFCS level I or II, and 88% had a spastic subtype of CP (of whom 49% unilateral and 51% bilateral spastic CP). Participants' characteristics are presented in SI 4.

Table I: Course over time for the total cohort of six participation domains of the Rotterdam Transition Profile by GMFCS level

	Leisure (social activities)				Transportation			
	N[a]	OR	95% CI	p	N[a]	OR	95% CI	p
Age	294	1.71	1.34 - 2.17	<0.001	299	1.57	1.34 - 1.83	<0.001
GMFCS III-V[b]	294	0.56	0.09 - 3.57	0.536	299	0.12	0.03 - 0.43	0.001
Age*GMFCS III-V[b]		NA	NA	NA	299	0.85	0.60 - 1.20	0.355

	Finances				Intimate Relationships			
	N[a]	OR	95% CI	p	N[a]	OR	95% CI	p
Age	388	1.56	1.40 - 1.73	<0.001	397	1.28	1.19 - 1.37	<0.001
GMFCS III-V[b]	388	5.05	1.29 - 19.77	0.020	397	0.29	0.14 - 0.62	0.001
Age*GMFCS III-V[b]	388	1.23	0.87 - 1.73	0.235	397	0.98	0.87 - 1.11	0.773

	Education and employment				Housing			
	N[a]	OR	95% CI	p	N[a]	OR	95% CI	p
Age	396	1.63	1.45 - 1.82	<0.001	400	1.47	1.37 - 1.58	<0.001
GMFCS III-V[b]	396	0.71	0.34 - 1.45	0.347	400	0.66	0.30 - 1.44	0.294
Age*GMFCS III-V[b]	396	0.78	0.66 - 0.93	0.005	400	0.81	0.72 - 0.91	<0.001

[a]N represents the total number of observations in the models. The number of observations varied due to study design and the domains leisure and transportation were added later throughout the development of the Rotterdam Transition Profile. [b] Reference category: GMFCS level I-II. OR, odds ratio; CI, confidence interval; GMFCS, Gross Motor Function Classification System; NA, not applicable (model is inappropriate for data).

Transition into adulthood

Figure 1 shows the distribution of development of autonomy in the six domains of participation, by age. For transportation the majority was autonomous from age 13 years onwards; for leisure, finances, intimate relationships and employment the majority was autonomous from age 18-22 years onwards; and for housing the majority was autonomous from 27 years of age. Overall, at over 27 years of age, in each of the participation domains, 90% of individuals with CP were autonomous, with the exception of intimate relationships, for which the proportion of autonomous individuals levelled off at slightly over 70%.

Longitudinal development of autonomy in participation

The GEE analyses (Figure 2) show that the proportions of autonomous individuals with CP increased with age; Table 1 presents the model parameters. In addition, for transportation and intimate relationships, lower proportions of individuals with GMFCS levels III-V were autonomous compared to those with GMFCS levels I-II (ORs for autonomy: 0.12, 95% confidence intervals (CI): 0.03-0.43 and OR: 0.29, 95% CI: 0.14-0.62, respectively), regardless of age, since interactions were not significant. This means, for example, that individuals with lower gross motor function have lower odds to organize their transportation autonomously compared to individuals with higher gross motor function. Notably, for finances, the proportions of autonomous individuals were higher for those with GMFCS levels III-V, (OR: 5.05, 95% CI: 1.29-19.77), regardless of age. Finally, for employment and housing, the development with age differed between individuals with GMFCS levels III-V versus GMFCS levels I-II, as indicated by significant interactions (Table 1).

Table 2: Proportions of autonomy in education and employment, finances, housing and intimate relationships in individuals with cerebral palsy (CP) and Dutch age-matched population

	Finances (independent life, phase 3)				Intimate Relationships (independent life, phase 3)			
Age (years)	15-19	20-24	25-29	30-34	18-19	20-24	25-29	30-34
CP (%)	21.9[a]	64.4[a]	93.3	97.9	20.8[c]	50.3	60.0	74.5
Dutch general population (%)	54.2	78.7	89.6	95.2[b]	50.6	78.6	88.2	91.8
Binominal Test (p)	<0.001	<0.001	0.768	0.668	<0.001	<0.001	<0.001	<0.001

	Housing (independent life, phase 3)	
Age (years)	20-24	25-29
CP (%)	32.7	63.3
Dutch general population (%)	50.1	83.5
Binominal Test (p)	<0.001	0.012

Number of observations at subsequent age intervals for CP: 147 (15-19 years), 162 (20-24 years), 30 (25-29 years), 47 (30-34 years). [a] Missing data for finances (15-19 years n=10, 20-24 years n=2) and intimate relationships (20-24 years n=3). [b] Age Dutch general population 30-32 years. [c] Number of observations at age 18-19 years for CP: 77.

From the late twenties onwards, the development of individuals with GMFCS levels III-V levelled off and, therefore, reached lower proportions of autonomy in employment and housing compared to those with GMFCS levels I-II; in their late twenties, on average 64% of individuals with GMFCS III-V were autonomous in employment and 65% in housing (Figure 2).

Comparison with the general population

A comparison was made between autonomy of individuals with CP and Dutch aged-matched references for three domains (Table II): finances, intimate relationships, and housing. Within the age range 15-24 years, lower proportions of individuals with CP were autonomous (according to the RTP) compared to reference data of the Dutch general population for these life areas. For individuals aged ≥25 years, no significant differences were found for finances, whereas for intimate relationships and housing the proportions of autonomous individuals with CP remained lower compared to the Dutch general population at this age.

DISCUSSION

This study describes the long-term development of autonomy in participation of individuals with CP without intellectual disability, from their youth into adulthood. With increasing age, more individuals with CP became autonomous in participation; specifically, within each of the domains (with the exception of intimate relationships) over 90% of the sample reached autonomy in their late twenties. Over the total age range, individuals with GMFCS levels III-V were less often autonomous in transportation, were more often economically independent (finances), and less often had experience with a sexual relationship (intimate relationships), compared to those with GMFCS levels I-II. In their late twenties, differences emerged between individuals with GMFCS levels I-II and GMFCS levels III-V for employment and housing. Individuals with CP appeared to lag behind compared to the age-matched Dutch population in finances, intimate relationships, and housing.

The increase of autonomy in participation with increasing age was expected based on the developmental concept of the RTP and the earlier longitudinal study that included part of the present sample.[4] Lower levels of gross motor function were related to less autonomy in transportation and active sexual relationships over the total age range, but not to financial independence; this is in line with a study conducted in Norway.[4,8] The latter finding regarding financial independence is likely due to regulations for disability benefits for adults with work limitations (age ≥18 years) in the Netherlands and Norway. For employment and housing we identified different patterns of development of autonomy for the subgroup with lower gross motor function compared to those with higher gross motor function. The proportions of autonomous individuals with GMFCS levels I-II continued to increase with increasing age to almost 100%, whereas a substantial proportion (± 35%) of individuals with GMFCS level III-V did not reach autonomy in employment and housing.Therefore, individuals with lower gross motor function are at risk not to achieve autonomy in transportation, intimate relationships, employment and housing. Rehabilitation professionals should be aware of this and adequately address development of autonomy in personalized treatment.

Compared to their Dutch peers, the total sample of individuals with CP aged ≤25 years was less often autonomous in finances, intimate relationships, and housing; this is in line with earlier studies.[4,23,19,24] The present results also show that, for intimate relationships and housing, individuals with CP continue to lag behind throughout their early thirties compared to their

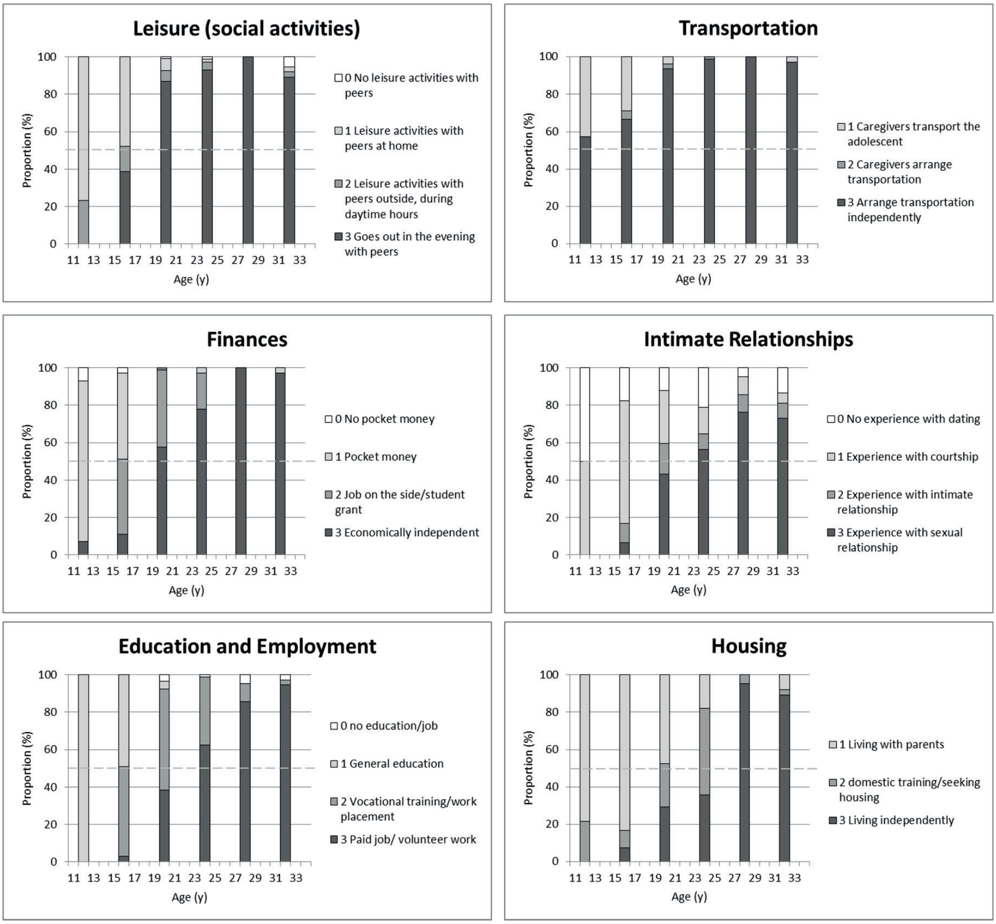

Figure 1: Development of autonomy in participation over time (range 12-34 years) for a cohort of young adults with CP and without intellectual disability presented in age categories of 4 years.

The dashed line indicates the 50% level.

typically developing peers, whereas for finances the differences compared with peers decrease above age 25 years; this decrease is probably also due to the system of disability benefits in the Netherlands. For the domain of employment, no reference data were compared as the RTP considers work participation including voluntary work, whereas Statistics Netherlands does not. For intimate relationships, the difference between individuals with CP and the reference data may be even larger since, for the references, experience in intimate relationships was only considered over the previous 12 months. For intimate relationships and housing, overall fewer individuals with CP reached autonomy, indicating that individuals with CP may benefit from specialized support in these areas. This is confirmed by a need for information and intervention on CP and sexuality, as expressed by youth with CP themselves, who reported that sexuality is scarcely discussed in rehabilitation treatment.[24] A specialized group program might help to address this need.[25] For housing, specialized support may include residential training for individuals with CP in their early twenties.

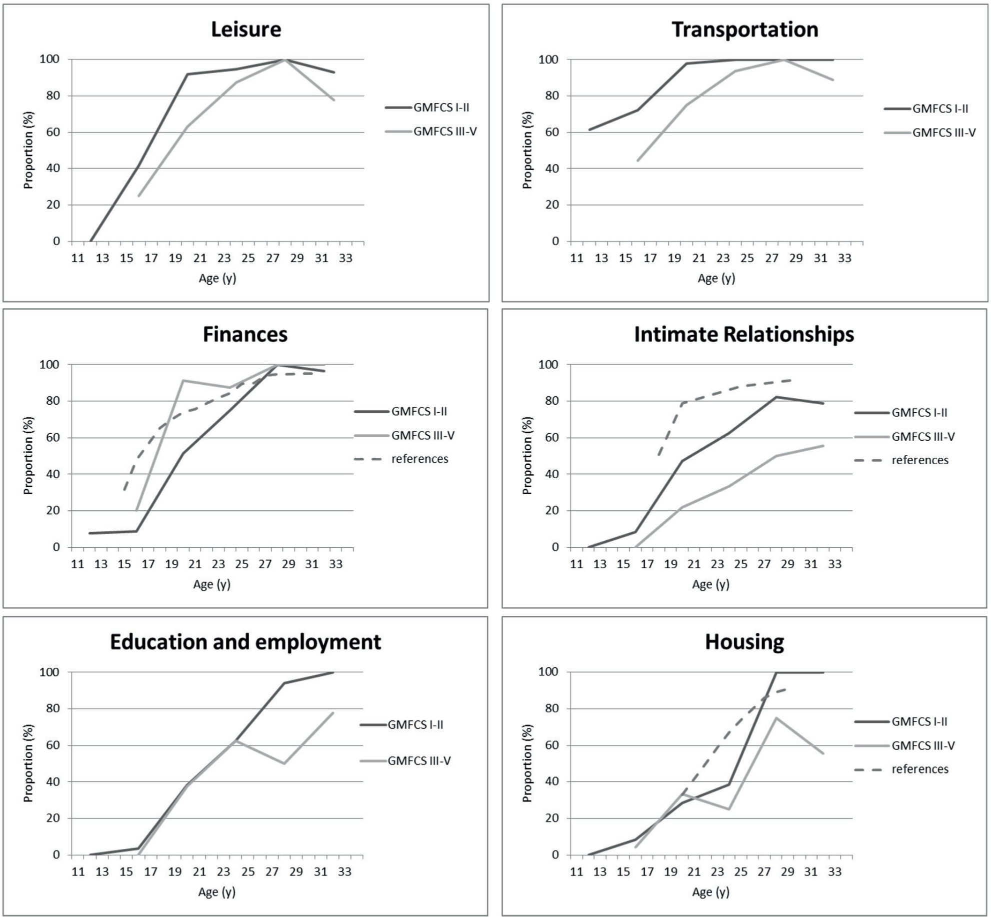

Figure 2: Proportions of individuals with cerebral palsy in phase 3 of the Rotterdam Transition Profile over time, and specified by Gross Motor Function Classification System (GMFCS) presented in age categories of 4 years.

Number of observations at subsequent age intervals for CP: 14 (12-14 years), 108 (15-18 years), 147 (19-22 years), 73 (23-26 years), 21 (27-30 years),37 (31-34 years). Due to missing data number of observations were for leisure (15-18 years n=45, 19-22 years n=107, 23-26 years n=72), transportation (15-18 years n=45, 19-22 years n=109), finances (15-18 years n=100, 19-22 years n=144, 23-26 years n=72), intimate relationships (19-22 years n=146, 23-26 years n=71) education and employment (19-22 years n=144, 23-26 years n=72). The dashed line indicates proportions of autonomy of the Dutch age-matched population; this added for visual estimation, whereas binomial tests are presented on total study population.

The RTP was used to assess autonomy, which we consider an aspect of participation. The RTP is easily administered (longitudinally) to evaluate the process of transition from adolescence to adulthood; the present results add to the evidence that the RTP can also capture changes with increasing age. In accordance with the definition of autonomy, the RTP covers two dimensions of autonomy, decisional autonomy (self-determination) and executional autonomy (independence).[2] Additional qualitative studies are required to further elucidate these specific dimensions of autonomy.

Strengths and Limitations

This is the first study to describe the development of autonomy in participation of individuals with CP without intellectual disability over a broad age range, from teens through the early thirties. Despite substantial dropout since baseline (SI 2), our rehabilitation-based sample seemed representative for the population as the distributions of sex, GMFCS level and type of CP were similar to individuals with intellectual disability in population-based studies. [26] In line with these distributions, we had lower numbers of participants with GMFCS levels III-V, increasing the uncertainty around odds ratio's comparing individuals with GMFCS levels III-V to those with levels I-II. Another limitation is that a substantial part (46%) of the sample was assessed cross-sectionally and, for those aged 12-16 years, only cross-sectional observations were available; therefore, these parts of the results are less robust. Since autonomy in participation is influenced by national legislation, for instance regarding social services, sheltered employment, and disability benefits, the present results should be interpreted with caution and estimated proportions may not be generalizable to other countries.

Clinical relevance and recommendations for future research

The present study offers insight into the development of autonomy of individuals with CP for several participation domains. The results show that individuals with lower gross motor function are at risk of not achieving autonomy; this should be addressed in rehabilitation care, especially regarding intimate relationships, employment and housing.[27,25] Future research may add knowledge concerning barriers and facilitators influencing the development of autonomy in different life areas for individuals with CP, as previously investigated for intimate relationships.[24,10] Since the present study focused on individuals without intellectual disability, future research may also examine the development of autonomy in participation and needs of those *with* an intellectual disability.

CONCLUSION

In this study, in most life areas, 90% of individuals with CP without intellectual disability reached autonomy in adult roles. Over the total age range, individuals with GMFCS levels III-V were less often autonomous in transportation and intimate relationships compared to those with GMFCS levels I-II. In the late twenties, differences between those with GMFCS levels I-II and levels III-V also emerged in employment and housing. Compared to the aged-matched general population, individuals with CP seem to lag behind in the development of autonomy in their teens until their early thirties. These results urge rehabilitation professionals to address the development of autonomy and help guide expectations, especially in individuals with lower gross motor function for intimate relationships, employment and housing.

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7

Childhood factors predict participation of young adults with cerebral palsy in domestic life and interpersonal relationships: a prospective cohort study

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ABSTRACT

Purpose: To determine childhood predictors of participation in domestic life and interpersonal relationships of young adults with cerebral palsy.

Materials and Methods: This 13-year follow-up of an existing cohort (baseline age 9-13 years) included 67 young adults with cerebral palsy (age 21-27 years). The Vineland Adaptive Behavior Scales and Life Habits questionnaire were used to assess attendance and difficulty in participation in domestic life and interpersonal relationships. Baseline factors were categorized according to the international classification of functioning, disability and health. Stepwise multiple linear regression analyses determined significant predictors ($p<0.05$).

Results: Lower manual ability, epilepsy, intellectual disability and lower motor capacity predicted decreased participation in domestic life and/or interpersonal relationships (explained variance R^2=67-87%), whereas no association was found with environmental and personal factors. Extending models with baseline fine motor skills, communication and interpersonal relationships increased R^2 to 79-90%.

Conclusions: Childhood factors account for 78-90% of the variation in young adult participation in domestic life and interpersonal relationships of individuals with cerebral palsy. Children with limited motor capacity, low manual ability, epilepsy or intellectual disability are at risk for restrictions in participation in young adulthood. Addressing communication and interpersonal relationships in pediatric rehabilitation might promote young adult participation.

INTRODUCTION

Cerebral palsy (CP) describes a group of permanent disorders of movement and posture, attributed to non-progressive disturbances in the developing fetal or infant brain, causing activity limitations.[1] The International Classification of Functioning, Disability and Health (ICF) defines participation as '*involvement in a life situation*' and describes its relation with an individual's health status, body functions & structures, ability to perform activities, and also with environmental and personal factors.[2] For activities and participation, the ICF describes the qualifier capacity as '*what one can do in a standardized environment*', and the qualifier performance as *'what one actually does in their current environment'* [2]. Since most children with CP now survive into adulthood, and young adults with CP are known to be restricted in their participation, insight is required to help early identification of individuals at increased risk of future restrictions in participation.[3-5]

Among young adults with CP, a large proportion experiences difficulty in participation, particularly in domestic life.[6] Moreover, for domestic life and interpersonal relationships, these proportions increase from age 16 years onward.[6] Participation in *domestic life* includes activities in/around the protected environment of one's home, e.g. preparing meals and doing housework.[2] Participation in *interpersonal relationships* includes socially appropriate interactions with others in various contexts, e.g. maintaining both formal and intimate relationships.[2] Because of the different contexts and different types of activities in these domains of participation, the predictors of these two domains are expected to vary. Therefore, and also because of increasing difficulties in both domains from teenage years into adulthood, these two are of particular interest.

Previously, we reported on cross-sectional associations between participation and CP-related characteristics, body functions and environmental and personal factors that were explored in youth and young adults with CP.[7-10] These (and other) studies demonstrated that individuals who were more severely functionally affected were more restricted in both their domestic life and interpersonal relationships.[7,9-11] In addition, these studies revealed that different factors are associated with either domestic life or interpersonal relationships. For example, for domestic life, adequate adaptations in the home environment were related to higher participation.[11] For interpersonal relationships, restricted participation was associated with having epilepsy and several environmental (e.g. less favorable attitudes of family and friends) and personal factors (e.g. behavior problems).[8,9,11] To enable clinicians to timely optimize treatment, longitudinal studies are needed that provide information on factors predicting future participation, in addition to the above-mentioned cross-sectional associations.

Until now, for individuals with CP, the predictors of participation have only been studied longitudinally among youth and over a relatively short period of time.[12,13] The baseline level of participation was shown to be the most important predictor of participation five years later.[12] Also, being more affected by CP (e.g. by having poorer walking ability or intellectual disability) and psychological problems in childhood predicted more limitations in participation in domestic life five years later.[12] For domestic life, CP-related characteristics explained a larger part of future participation compared to interpersonal relationships.[12] Furthermore, environmental factors (e.g. parental stress) predicted poorer future participation in interpersonal relationships, but not in domestic life.[12,13] However, it remains unclear whether childhood factors also predict participation in young adulthood.

Insight into childhood factors predicting future participation as young adults may identify: i) which individuals with CP are at risk for restricted participation, and ii) provide information on modifiable factors that can be addressed in pediatric rehabilitation. Therefore, this study aimed to determine whether childhood factors predict participation in domestic life and in interpersonal relationships of young adults with CP.

MATERIAL AND METHODS

Design

This study describes the 13-year follow-up of the PERRIN (Pediatric Rehabilitation Research In the Netherlands) 9-16 cohort, with previous yearly assessments over the course of 3 years.[9]

Participants

At baseline, 244 children with CP who were 9, 11 or 13 years of age were identified by rehabilitation centers, special education institutions for physically and mentally disabled children, and outpatient clinics of rehabilitation medicine departments in the northwest region of the Netherlands. These children and their parents were sent an information letter about the 3-year longitudinal study and invited to participate. Finally, 110 children and their parents returned the informed consent form and participated in the PERRIN 9-16 cohort. The study was approved by all regional medical ethics committees.

Participants of the PERRIN 9-16 cohort (*n*=110) were invited for a 13-year follow-up (PERRIN DECADE) at age 21-27 years, with the exception of one deceased participant and another three were excluded since they had a diagnosis other than CP that affected their motor functioning. Two mailings of information letters and a telephone call were carried out, and in case of no response, consecutively a telephone call or additional mailing was sent. In brief, participants had a clinical diagnosis of CP without additional disorders affecting motor functioning, and participants and their parents or caregivers were able to participate in face-to-face interviews in Dutch. The PERRIN DECADE study was approved by the Medical Ethical Committee of the VU University Medical Center, Amsterdam.

Procedure

At the 13-year follow-up, participants and/or their caregivers (caregivers were only present for interviews with individuals with intellectual disability; ID) were interviewed regarding participation in interpersonal relationships and domestic life at home or another location they selected. The Vineland Adaptive Behavior Scale second edition survey version (Vine-II) and the Assessment of Life Habits 3.1 (Life-H) were used, which evaluate different constructs of participation. Additionally, at 13-year follow-up an online questionnaire regarding the participant's living and civic status was completed by participants or, in case the participant had intellectual disability, their caregiver. Questions addressed participants' housing situation, housing type, and intimate relationships. Baseline factors were previously collected from the child's caregiver, using various instruments and questionnaires (see below).

Materials and instruments

Domestic life and interpersonal relationships in young adulthood

Vineland Adaptive Behavior Scale second edition survey version (Vine-II)

The Vine-II assesses whether or not activities are performed in daily life areas, which addresses an aspect of attendance of participation. Therefore, Vine-II scores are further indicated as participation attendance. The Vine-II covers domains of communication, daily activity, socialization and motor skills. For the present study, the participation subdomains of 'domestic daily living skills' and 'interpersonal relationships' were used. Items were scored as never performed (0), sometimes or partially performed (1), or usually or habitually performed (2). Performance could include the use of assistive devices or adaptations, if individuals used these in their usual functioning. The Vine-II has high intra-rater reliability and moderate inter-rater reliability and is validated in healthy children and adults, individuals with ID, and children with hearing or visual impairment [14]. Individuals with a score lower than one standard deviation (SD) below the mean reference value were considered to function below an adequate level according to their age.

Assessment of Life Habits 3.1 (Life-H)

The Life-H questionnaire 3.1 assesses participation performance, further qualified by experienced difficulty and assistance required, with performance in 12 domains of daily activities and social roles. For the present study, the domain scores of 'housing 'and 'interpersonal relationships' were used and are further reported as difficulty in participation in these domains. For each applicable item, difficulty was scored as 'no difficulty', 'some difficulty', 'accomplished by a proxy' or 'not accomplished'. Assistance was scored as 'no assistance', 'use of assistive device', 'adaptation' and/or 'with human assistance' (dependent functioning). From both scores, an item score was derived, from which a sum score of applicable items was calculated for each domain (range 0-10).[15] A domain score < 8.89 reflects participation with difficulty. The Life-H was developed for individuals with disabilities, has good intra- and interrater reliability, and good discriminant and construct validity in adults with spinal cord injury and stroke.[16,17]

Baseline factors

Factors assessed at baseline were categorized according to the ICF components addressing health condition, body functions and structures, motor capacity, activity & participation, environmental factors and personal factors.

Health condition included the CP-related classifications Gross Motor Function Classification System (GMFCS), Manual Ability Classification System (MACS) and CP subtype. GMFCS and MACS are classifications for functional severity ranging from I (highest functional level) to V.[18,19] Since no baseline data of MACS level were available, the 3-year follow-up assessment was used. To reduce the number of independent variables, the GMFCS level was subdivided in three categories: levels I and II, III and IV, and level V. MACS level was dichotomized: levels I and II versus levels III-V. CP subtype was categorized in spastic (both unilateral and bilateral) or non-spastic CP (ataxic, dyskinetic or mixed subtype).[20]

Body functions and structures included ID (following a special education program for children with ID, no/yes), epilepsy (more than one seizure during the previous two years or using antiepileptic drugs, no/yes), visual impairment (use of visual aids, no/yes), hearing impairment (use of a hearing device, no/yes), and speech problems (using the item 'speech problems' in the Child Behavior Checklist (CBCL), no/somewhat or very true).[21]

Motor capacity was assessed with the 66-item Gross Motor Function Measure-66 (GMFM-66), a standardized observational instrument developed to assess gross motor skills of children with CP in a standardized test situation. The items were scored on a 4-point scale and analyzed with the Gross Motor Ability Estimator to obtain a ratio scale GMFM-66 score.[22]

Activity & participation further included the baseline performance of motor skills (gross and fine), communication (receptive, expressive and written), daily living skills (personal, domestic and community) and socialization (interpersonal relationships, play & leisure and coping skills), assessed with the corresponding subdomains of the Vineland Adaptive Behavior Scale (VABS) survey. The VABS is the preceding version of the Vine-II, which was not available at the time of the baseline assessments. VABS items are scored as never performed (0), sometimes or partially performed (1), or usually or habitually performed (2), which are summed for each subdomain. The VABS is a reliable and valid instrument to assess activity and participation performance of children by means of a semi-structured interview and is validated for use in individuals with ID and in children with hearing or visual impairments.[23]

Environmental factors included housing type (regular/adjusted), the child's type of education (regular/special), the number of siblings (0 or 1/≥2), parental level of education (low/intermediate: upper secondary vocational education and lower, or high: secondary non-vocational higher education and university), marital status of parents (single/married or with partner), and parental stress and support. Parental stress and support were measured with a questionnaire based on the Dutch version of Moos' Life Stressors and Social Resources Inventory. Items were scored on a 4-point scale, with higher scores indicating more stress and less social resources. Mean domain scores were calculated for relational stress and social resources (items on interpersonal relationships) and situational stress and resources (items on financial and material resources, and life events).[24]

Personal factors included gender, age, nationality (Dutch or other), behavioral problems, and perceived self-competence. Behavioral problems were assessed with the six domains of the CBCL, depression, anxiety, withdrawal, somatization, delinquency and aggression. Items reflect behavioral problems, and are scored as 0=not applicable, 1=somewhat applicable, 2=applicable. Two sum scores were calculated: 1) for internalizing behavior (depression, anxiety, withdrawal, somatization) and 2) for externalizing behavior (delinquency, aggression). The CBCL has good reliability in children with developmental delays.[21] Self-competence was assessed with Harter's Social Perception Profile for children (SPPC), adjusted for use in children with CP. The SPPC has six scales: scholastic competence, social acceptance, athletic competence, physical appearance, global self-worth and motor competence. Each scale score is the mean of items that are scored on a four-point scale.[25,26]

Statistical analysis

Starting at baseline, from the PERRIN 9-16 cohort, four annual observations were available for motor capacity (GMFM-66) and activity & participation (VABS). To reduce the influence of measurement error, all four observations were used to model baseline scores that were used in the analyses. To model baseline scores, linear mixed model analyses were used with age as covariate, a random intercept to allow individual estimation of the baseline value, and (if applicable) a random slope for age. For baseline factors and for participant characteristics at the 13-year follow-up, descriptive statistics were computed.

To determine which baseline factors best predict participation in domestic life and interpersonal relationships at 13-year follow-up, stepwise multiple linear regression analyses were performed. Dependent variables were the (sub)domain scores of domestic life and interpersonal relationships of the Vine-II and Life-H. For each ICF component, a forward selection procedure was conducted until no additional factor contributed with a cut-off p-value <0.1. Then, final prediction models were determined, again using a forward selection procedure (cut-off p-value <0.05), including only the selected factors of the ICF components CP-related characteristics, body functions, motor capacity, and environmental and personal factors. In an additional step, we examined whether the childhood level of activity and participation performance provided additional value to these final prediction models. Therefore, these extended models were determined by adding childhood activity and participation factors (one by one) to the final prediction models, to investigate whether possible modifiable factors could be identified. If more than one factor made a significant contribution, the strongest ones were selected using a forward selection procedure (cut-off p-value <0.05).

To check for potential influence of dropouts the baseline distribution of sex, ID, GMFCS level and type of CP of individuals who dropped out (n=40) were compared to those included (n=67) using a chi-square test. All analyses were done using SPSS for Windows (SPSS 22, IBM SPSS Statistics, Armonk, NY, USA).

RESULTS

Participants

Of the 106 invited PERRIN 9-16 participants (existing cohort), 22 declined and 17 did not respond. Thus, the 13-year follow-up included 67 young adults with CP aged 21-27 years. Table 1 presents the participants' characteristics at the 13-year follow-up and characteristics of individuals who dropped out (n=40). Dropout was not selective regarding sex, ID, CP subtype and GMFCS level at baseline.

Participation in domestic life and interpersonal relationships

On average, for domestic life, young adults with CP scored 30.8 (SD 17.5) on the Vine-II, with 73% of individuals performing below an adequate level according to their age. Difficulty was experienced by 66% of individuals, with a mean score of 7.2 (SD 2.4) on the Life-H. For interpersonal relationships, young adults with CP on average scored 65.5 (SD 14.9) on the Vine-II, with 64% of individuals performing below an adequate level according to their age. Difficulty was experienced by 33% of individuals, with a mean score of 8.6 (2.0) on the Life-H.

Included factors

Table 2 presents the (modeled) baseline factors. Within each ICF component, the significant predictors are presented for each of the four participation outcome measures.

Predictors domestic life

The final prediction models for attendance (Vine-II) and difficulty (Life-H) in participation in domestic life are presented in table 3 (explained variance 87% and 79%, respectively). Having ID

Table 1: Characteristics of participants at 13-year follow-up and of non-participants

	Participants	**Non-participants**
	***n* (%)**	***n* (%)**
Gender	45/22 (67/33)	22/18 (55/45)
Male/female		
Age in years: mean (SD); min-max	24.6 (1.6); 21.6 to 27.4	na
GMFCS[a]		
I	30 (45)	19 (48)
II	7 (10)	7 (18)
III	8 (12)	5 (13)
IV	9 (13)	4 (10)
V	13 (19)	5 (13)
MACS[b]		
I	26 (39)	13 (45)
II	22 (33)	11 (38)
III	7 (10)	2 (7)
IV	7 (10)	3 (10)
V	5 (7)	0
		missing: 11
Type of CP		
Spastic	52 (78)	34 (85)
Unilateral	21	16
Bilateral	31	18
Dyskinetic	3 (5)	1 (3)
Ataxic	3 (5)	1 (3)
Mixed	9 (13)	4 (10)
ID		
No/yes	46/21 (69/31)	29/11 (73/28)
Housing situation		
with parents	36 (55)	na
alone	17 (26)	
with partner	2 (3)	
other	10 (15)	
	missing[c]: 2	
Housing type		
Regular housing	34 (52)	na
Adjusted housing	17 (26)	
Assisted housing	14 (22)	
	missing[c]: 2	
Ever in a romantic relationship		
Yes/no	34/28 (55/45)	na
	missing[c]: 5	

[a] GMFCS data were assessed at baseline and considered constant for analysis

[b] MACS data were assessed at the 3-year follow-up or at the 13-year follow-up in case of missing values (*n*=3) and considered constant for analysis

[c] Missing: 2 participants did not complete the online questionnaire. Additionally, 3 participants chose not to answer question on romantic relationships

na: not available, since this data was collected at the 13-year follow-up

SD: standard deviation

GMFCS: Gross Motor Function Classification System

MACS: Manual Ability Classification System

CP: cerebral palsy

ID: intellectual disability

or a lower GMFM-66 score predicted lower attendance and more difficulty in participation in domestic life in young adulthood. Compared to participants in MACS I-II, individuals in MACS III-V had lower future participation attendance (but not more difficulty) in domestic life. Extending the attendance model of domestic life with baseline activity and participation did not improve the model. For difficulty, the extended model included the baseline performance of fine motor skills and receptive communication; these factors added 7.4% of the explained variance compared to the prediction model without activity and participation.

Predictors interpersonal relationships

The final prediction models for attendance (Vine-II) and difficulty (Life-H) in participation in interpersonal relationships are also presented in table 3 (explained variance 74% and 67%, respectively). Having epilepsy or ID predicted lower attendance and more difficulty in participation in interpersonal relationships in young adulthood. Those in MACS III-IV compared to individuals in MACS I-II had lower future participation attendance (but not difficulty) in interpersonal relationships. In addition, a lower baseline value of the GMFM-66 predicted more difficulty.

For attendance in interpersonal relationships the extended model included baseline expressive communication, and for difficulty the extended model included baseline interpersonal relationships; both these latter factors added 16.5% and 11.9%, respectively, of the explained variance compared to the prediction models without activity & participation. In these extended models, epilepsy (for attendance) or ID (for difficulty) were no longer significant predictors.

DISCUSSION

This study explored childhood factors of individuals with CP that may predict future participation in domestic life and interpersonal relationships in adulthood. It was found that childhood factors explained a large part of the variance in young adult participation, i.e. up to 90%. Also, individuals with low motor capacity, low manual ability and ID were at increased risk for lower levels of future participation in domestic life. Similarly, future participation in interpersonal relationships was lower for these same individuals and, additionally, for those with epilepsy. However, in these models, no environmental or personal factors were identified as significant predictors. Extending the models with childhood activity and participation levels substantially improved the models for future interpersonal relationships.

Risk factors

The finding that CP-related factors and body functions predict future participation is in accordance with Dang et al. who found that, over a 5-year period, impairment (including level of gross motor function, level of manual ability, ID, epilepsy and communication impairments) predicted participation in the domestic life of adolescents with CP.[12] Similarly, we previously found that epilepsy and ID were longitudinally associated with the development of social participation in all PERRIN cohorts, covering a broad age range.[27] The CP-related factors and body functions that we identified as predictors for future participation indicate that more severely affected individuals are at risk of reaching lower levels of participation as young adults. Epilepsy, ID, low manual ability and low motor capacity are often interrelated in CP, i.e. Individuals with less favorable motor function more often have ID and or epilepsy.[28]

Table 2: Possible predictive factors by ICF component at baseline.

		Domestic life		Interpersonal relationships	
	Total *n*=67, age at baseline: 9-13 years	Attendance (Vine-II)	Difficulty (Life-H)	Attendance (Vine-II)	Difficulty (Life-H)
CP-related characteristics (health condition)	*n* (%)	Selected predictors of cluster, β (SE); R^2			
GMFCS in 3 categories					
I + II	37 (55)	ref category	ref category	ref category	ref category
III + IV	17 (25)	-16.66 (2.69) ***	-1.80 (0.39) ***	-4.99 (3.31)	-0.56 (0.42)
V	13 (19)	-27.62 (4.12) ***	-5.26 (0.43) ***	-19.45 (5.07) ***	-3.9 (0.49) ***
MACS in 2 categories					
I + II	48 (72)	ref category		ref category	ref category
III + IV + V	19 (28)	-11.66 (3.46) ***		-8.45 (4.26) *	0.92 (0.45) **
Type of CP (spastic/other)	52/15 (78/22)				
		R^2: 76%	R^2: 70%	R^2: 50%	R^2: 51%
Body functions and structures	*n* (%)	Selected predictors of cluster, β (SE); R^2			
ID (no/yes)	46/21 (69/31)	-17.88 (3.81) ***	-2.92 (0.52) ***	-18.50 (2.74) ***	-1.97 (0.37) ***
Epilepsy (no/yes)	61/6 (91/9)	-11.44 (5.27) **	-1.42 (0.74) *	-17.30 (4.45) ***	-3.19 (0.59) ***
Visual impairment (no/yes)	53/14 (79/21)	-15.00 (4.07) ***	-1.35 (0.57); 0.021**		
Hearing impairment (no/yes)	64/3 (96/4)				
Speech problems (item 79 of CBCL) (no/yes)	35/31 (52/46) missing: 1 (1)	R^2: 62%	R^2: 61%	R^2: 59%	R^2: 58%
Motor capacity	mean (SD); min-max	Selected predictors of cluster, β (SE); R^2			
Gross motor capacity (GMFM-66) *(66 items, range 0-100)*	64.73 (28.28); 3.66 to 99.45	0.55 (0.04) ***	0.07 (0.01) ***	0.35 (0.05) ***	0.05 (0.01) ***
		R^2: 79.7%	R^2: 71%	R^2: 44.3%	R^2: 40.6%

Table 2: Continued

		Domestic life		Interpersonal relationships	
	Total *n*=67, age at baseline: 9-13 years	Attendance (Vine-II)	Difficulty (Life-H)	Attendance (Vine-II)	Difficulty (Life-H)
Activities and participation	mean (SD); min-max	Selected predictors of cluster, β (SE); R^2			
VABS Motor skills					
- Gross *(20 items, range 0-40)*	24.23 (12.45); 2.07 to 39.23	0.82 (0.14) ***	0.06 (0.02) ***		
- Fine *(16 items, range 0-32)*	23.73 (10.23); 0.10 to 31.70	0.62 (0.17) ***	0.14 (0.03) ***	1.13 (0.11) ***	0.14 (0.02) ***
		R^2: 81%	R^2: 77%	R^2: 60%	R^2: 50.4%
VABS Communication					
- Receptive *(13 items, range 0-26)*	24.42 (3.94); 2.13 to 25.94		0.19 (0.05) ***		0.31 (0.04) ***
- Expressive *(31 items, range 0-62)*	53.24 (14.03); 2.45 to 60.50			0.83 (0.07) ***	
- Written *(23 items, range: 0-46)*	22.66 (12.84); 0.06 to 40.39	1.14 (0.09) ***	0.12 (0.02) ***	0.23 (0.07) ***	0.05 (0.01) ***
		R^2: 70%	R^2: 70%	R^2: 87%	R^2: 71%
VABS Daily living skills					
- Personal *(39 items, range 0-78)*	55.36 (21.31); -0.30 to 75.88	0.75 (0.04) ***	0.10 (0.03) ***		0.07 (0.01) ***
- Domestic *(21 items, range 0-42)*	14.34 (6.98); 0.61 to 25.02				
- Community *(32 items, range 0-64)*	30.55 (13.99); -0.55 to 47.19		0.04 (0.02) **	0.91 (0.07) ***	
		R^2: 82%	R^2: 82%	R^2: 73%	R^2: 57%

Table 2: Continued

		Domestic life		Interpersonal relationships	
	Total *n*=67, age at baseline: 9-13 years	Attendance (Vine-II)	Difficulty (Life-H)	Attendance (Vine-II)	Difficulty (Life-H)
VABS Socialization					
- Interpersonal relationships *(28 items, range: 0-56)*	42.08 (7.79); 11.21 to 49.01			0.63 (0.35) *	0.10 (0.05) **
- Play and leisure *(20 items, range: 0-40)*	29.44 (7.14); 1.98 to 36.14		0.14 (0.06) **	0.74 (0.34) **	0.13 (0.05) **
- Coping (*18 items, range: 0-36)*	26.93 (8.70); 0.72 to 34.75	1.28 (0.19) ***	0.10 (0.05) **	0.40 (0.23) *	
		R^2: 40%	R^2: 59%	R^2: 78%	R^2:72%
Environmental characteristics	*n* (%)	Selected predictors of cluster, β (SE); R^2			
Housing type (non-adjusted/adjusted home)	38/29 (57/43)	-15.25 (3.1) ***	-2.22 (0.44) ***	-10.82 (3.34) ***	-0.90 (0.48) **
Educational type child (regular/special education)	31/36 (46/54)	-16.57 (3.12) ***	-2.08 (0.44) ***	-9.52 (3.28) ***	-1.27 (0.48) **
Parental education (low/high)	34/23 (51/34) missing: 10 (15)				
Marital status parents (single/living with partner)	6/61 (9/91)				
Siblings (0 and 1/2 or more)	12/55 (18/82)				
Life Stressors and Resources: mean (SD); min-max					
- Relational stress/support *(7 items, range 1-4)*	1.7 (0.4); 1 to 2.6			-7.78 (3.99) *	
- Situational stress/support *(13 items, range 1-4)*	1.7 (0.4); 1 to 2.8			6.47 (3.53) *	
		R^2: 60%	R^2: 58%	R^2: 41%	R^2: 24%
Personal characteristics	*n* (%)	Selected predictors of cluster, β (SE); R^2			
Gender (Male/female)	45/22 (67/33)				no significant factors
Ethnicity (Dutch/ other)	62/5 (93/7)				
Behavior problems (CBCL): mean (SD); min-max					
- Internalizing behavior problem *(32 items, range 0-64)*	9.4 (7.5); 0 to 34.0				

Table 2: Continued

		Domestic life		Interpersonal relationships	
	Total *n*=67, age at baseline: 9-13 years	Attendance (Vine-II)	Difficulty (Life-H)	Attendance (Vine-II)	Difficulty (Life-H)
Personal characteristics	*n* (%)	Selected predictors of cluster, β (SE); R^2			
- Externalizing behavior problem *(33 items, range 0-66)*	8.3 (6.8); 0 to 40.0	-0.70 (0.28) **			
	missing: 1				
Self-competence (SPPC): mean (SD); min-max					
- scholastic competence *(6 items, range 1-4)*	2.9 (0.7); 1.1 to 4.0				
- social acceptance *(6 items, range 1-4)*	3.0 (0.7); 1.3 to 4.0				
- athletic competence *(6 items, range 1-4)*	2.6 (0.6); 1.1 to 3.6		-0.86 (0.39) **		
- physical appearance *(6 items, range 1-4)*	3.2 (0.7); 1.3 to 4.0	6.79 (4.03) *			
- global self-worth *(6 items, range 1-4)*	3.3 (0.6); 1.4 to 4.0	-10.42 (4.15) **		-1.39 (0.78) *	
- motor competence *(6 items, range 1-4)*	2.8 (0.6); 1.8 to 4.0	6.21 (2.40) **	1.17 (0.39) **	1.64 (0.79) **	
	missing: 19				
		R^2: 27%	R^2:18%	R^2: 12%	

* p-value <0.1
** p-value <0.05
*** p-value <0.01
Vine-II: Vineland Adaptive Behavior Scale second edition survey version
Life-H: Assessment of Life Habits 3.1
β: Regression coefficient
SE: standard error
R^2: explained variance
CP: cerebral palsy

GMFCS: Gross Motor Function Classification System
MACS: Manual Ability Classification System
SD: standard deviation
ID: intellectual disability
CBCL: Child Behavior Checklist
GMFM: Gross Motor Function Measure
SPPC: Harter's Social Perception Profile for children
VABS: Vineland Adaptive Behavior Scale

Nevertheless, a diversity of combinations of these factors occur in CP, and as we found them as independent predictors of future participation, they should also be considered separately [29]. Those with poorer gross and fine motor function abilities in childhood are likely to continue to experience more motor limitations in adulthood,[30] which is associated to lower levels of participation. In addition, their development of new participation skills may be more challenging, since we know from another study that those with lower gross motor function had more difficulty and needed assistance in participation as they develop into adult roles.[6] Finally, individuals with ID are known to show less favorable development of participation, with development stabilizing at relatively low levels during childhood.[31] Therefore, screening children with CP for low motor capacity, low manual ability, ID and epilepsy may help the timely identification of those at risk for lower participation in future domestic life and/or interpersonal relationships, who may benefit from early support or treatment in a personalized rehabilitation program to develop daily activities and participation in domestic life and interpersonal relationships.

Environmental & personal factors

In contrast with earlier work among adolescents with CP,[12] the present study identified no environmental or personal factors as predictors of participation on the long term. This was in particular unexpected for personal factors, since Dang et al. found that psychological problems were predictors of future participation and we assessed behavioral problems in a similar way to their study.[12,32] It is possible that behavior problems are predictive over a shorter period of time (e.g. 5 years), but do not predict participation on the long term, because they are subject to change over longer time periods. This might be explained by an earlier study (using the present cohort) that showed that behavior problems were observed in childhood but diminished during adolescence.[33] This positive development of personal factors with age might explain why childhood behavior problems do not affect adult functioning. Furthermore, the present study found that childhood environmental factors did not predict future participation, while previous studies in childhood showed a strong cross-sectional association between environmental factors (i.e. physical home environment, attitudes of classmates and social support) and participation.[11,12] A possible explanation for this might be that, in the present study, environmental factors were examined in less detail compared to the study of Colver et al. In the present study, the childhood environmental factors were found to be predictors in the separate ICF component analyses but did not reach significance in the final models (in which factors of all ICF components were combined). This can be understood when considering that, in accordance with adult roles, the environment of young adults with CP may have changed drastically compared to childhood. Thus, although the environment and the person were previously associated with the current level, or were predictive of the short-term future participation of children, our results show that they do not seem to predict long-term participation in addition to epilepsy, ID, manual ability and motor capacity, within a sample of individuals with CP with a broad variety of severity levels (reflective of the population of individuals with CP). To confirm this hypothesis, future studies need to examine more environmental factors and study these in more detail (e.g. attitudes of social environment and received treatment) in order to determine whether these are predictive of young adult participation. We additionally advise to consider more homogenous subgroups of CP (e.g. exclusively individuals without ID), since the large variance explained by CP-related factors may overrule that of personal and environmental factors.

Table 3: Final prediction models

	Domestic life						**Interpersonal relationships**					
	Attendance (Vine-II)			**Difficulty (Life-H)**			**Attendance (Vine-II)**			**Difficulty (Life-H)**		
Prediction models	β (SE)	St β	p-value	β (SE)	St β	p-value	β (SE)	St β	p-value	β (SE)	St β	p-value
MACS III-V compared to I-II	-8.49 (2.59)	-0.22	0.002	-	-	-	-12.94 (2.45)	-0.40	<0.001	-	-	-
ID	-10.54 (2.23)	-0.28	<0.001	-1.86 (0.38)	-0.36	<0.001	-13.07 (2.52)	-0.41	<0.001	-1.02 (0.41)	-0.24	0.016
Epilepsy	-	-	-	-	-	-	-17.29 (3.73)	-0.33	<0.001	-3.07 (0.54)	-0.45	<0.001
GMFM	0.35 (0.05)	0.56	<0.001	0.05 (0.01)	0.62	<0.001	-	-	-	0.03 (0.01)	0.37	<0.001
R^2	86.5%			78.9%			73.5%			66.6%		
Extended models	β (SE)	St β	p-value	β (SE)	St β	p-value	β (SE)	St β	p-value	β (SE)	St β	p-value
MACS III-V compared to I-II	-8.49 (2.59)	-0.22	0.002	-	-	-	-5.86 (1.60)	-0.18	0.001	-	-	-
ID	-10.54 (2.23)	-0.28	<0.001	-1.44 (0.33)	-0.27	<0.001	-5.45 (1.66)	-0.17	0.002	-0.47 (0.34)	-0.11	0.180
Epilepsy	-	-	-	-	-	-	-4.34 (2.53)	-0.08	0.091	-1.58 (0.50)	-0.23	0.002
GMFM -66	0.35 (0.05)	0.56	<0.001	0.03 (0.01)	0.35	<0.001	-	-	-	0.02 (0.01)	0.23	0.005
VABS fine motor skills	-	-	-	0.07 (0.02)	0.32	0.002	-	-	-	-	-	-
VABS receptive communication	-	-	-	0.10 (0.04)	0.17	0.010	-	-	-	-	-	-
VABS expressive communication	-	-	-	-	-	-	0.72 (0.07)	0.68	<0.001	-	-	-
VABS interpersonal relationships	-	-	-	-	-	-	-	-	-	0.13 (0.02)	0.61	<0.001
R^2	86.5%			86.3%			90.0%			78.5%		

Vine-II: Vineland Adaptive Behavior Scale second edition survey version
Life-H: Assessment of Life Habits 3.1
β: regression coefficient
St β: standardized regression coefficient
SE: standard error
R^2: explained variance

MACS: Manual Ability Classification System
ID: intellectual disability
GMFM-66: Gross Motor Function Measure
VABS: Vineland Adaptive Behavior Scale

Effects of childhood activity and participation level

For difficulty in domestic life, childhood fine motor and communication skills improved the model slightly. For interpersonal relationships, childhood levels of either expressive communication or interpersonal relationships improved both models substantially, indicating that better social skills in childhood are important for young adult participation. It can be understood that communication and interpersonal relationships are related from the importance of communication skills in interacting with others,[34] and the association of communication skills with relationships formed at school.[35] Professionals should be alert to appropriate functioning in these domains, particularly for children with epilepsy, intellectual disability, low manual ability, or low motor capacity who are at risk of lower levels of adult participation in interpersonal relationships.

Similarities and differences between participation domains

In this study, similar factors were identified as predictors of future participation for the two studied aspects of participation: attendance and difficulty. Differences were observed between the predictors of participation in domestic life and interpersonal relationships. First, in addition to motor capacity, manual ability and ID, that predicted both domestic life and interpersonal relationships, epilepsy only predicted participation in interpersonal relationships. This factor had a strong predictive value, albeit our sample included few individuals with epilepsy compared to the proportion observed in other CP populations.[36] Associations between epilepsy and interpersonal relationships were also found in a previous study based on the current cohort, as well as in other studies.[37-39] Individuals with epilepsy may experience participation problems in more complex environments, in contrast with the familiar home environment, which is where participation in interpersonal relationships takes place. These problems might be due to increased reticence about going out because of possible seizures, or to practical restrictions, e.g. related to traveling alone. Clinicians could pay special attention to youth with epilepsy regarding experienced obstacles in their participation in interpersonal relationships and take these into account in their rehabilitation treatment. Second, we found that factors regarding motor functioning (e.g. gross motor capacity, manual ability and fine motor skills) predicted participation in domestic life for a larger part than interpersonal relationships, which is in line with Dang et al.[12] This may also be understood from the different constructs of participation in domestic life and interpersonal relationships. Participation in domestic life includes mobility in the home environment and household tasks which presumably have a larger physical component. In conclusion, risk factors and possible modifiable factors differ between the participation domains of domestic life and interpersonal relationships, which suggests the need for individualized goal setting and rehabilitation care to optimize young adult participation.

Strengths and limitations

A strength of this study is the long follow-up (13 years), which allowed to determine early predictors of participation on the long term. Also, despite this long time interval, 63% of the baseline sample was included in the present follow-up. Nevertheless, in view of the relatively small sample size and the large number of childhood factors, we chose to use a forward-stepwise analysis. This approach provided additional insight into the strength of the associations of factors in the different ICF components with the outcomes. The present results and interpretations focused on the strongest childhood activity and participation factors only, although several subdomains were strongly associated with the outcomes. The results categorized by ICF domain of the activity and participation component can provide additional insight into these other associations (table 2). Finally, it should be noted that our analyses cannot ascertain causal relationships, and intervention studies are needed to determine whether rehabilitation treatment aimed at improving motor capacity, activity and participation in childhood indeed results in improved participation as a young adult.

CONCLUSIONS

In this study, childhood factors accounted for 78-90% of the variation in young adult participation in domestic life and interpersonal relationships. For the most part, this was explained by CP-related factors and body functions, whereas environmental and personal factors in childhood did not predict future participation as young adults. Children with CP with limited motor capacity, manual ability, epilepsy or ID are at risk for future participation restrictions in domestic life or interpersonal relationships in young adulthood. Addressing communication skills and interpersonal relationships in childhood rehabilitation may contribute to improving participation later in life.

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8

Teenage predictors for future participation of adults with cerebral palsy in domestic life and interpersonal relationships: a 13-year follow-up study

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ABSTRACT

Background: Adults with cerebral palsy (CP) may experience problems with participation in domestic life and interpersonal relationships.

Aims: To identify teenage predictors of adult participation in domestic life and interpersonal relationships.

Methods and procedures: This 13-year follow-up of the PERRIN 16-24 cohort included 53 adults with CP without intellectual disability [current age 31.7 (SD=1.4) years]. Participation performance was assessed as attendance (Vineland Adaptive Behavior Scales), and difficulty/assistance with participation (Life Habits questionnaire). 56 teenage factors were categorized in ICF components. Stepwise multiple linear regression analyses explored predictors of participation.

Outcomes and results: Lower gross motor capacity, following special education, having protective parents and a rigid personality predicted less participation in domestic life. Having rejective parents, receiving little daily support, having a socially avoidant personality or coping style and the male gender predicted less participation in interpersonal relationships. Lower activity and participation levels as a teenager predicted less participation in both domestic life and interpersonal relationships of adults with CP.

Conclusions and implications: Environmental and personal factors, gross motor capacity and teenage participation were predictors of participation of adults with CP. These factors help identify subgroups at risk for suboptimal adult participation and provide targets for rehabilitation.

INTRODUCTION

Cerebral palsy (CP) describes *'a group of disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain …'*.[1] Individuals with CP form the largest diagnosis group in pediatric rehabilitation. With improved life expectancy, adults with CP make up a large proportion of the population of individuals with CP. Adults with CP often experience challenges with daily activities and involvement in life situations; the latter is defined by the World Health Organization as 'participation'.[2] The International Classification of Functioning and Health (ICF) states that participation performance can be qualified by 'experienced difficulty' and 'needed assistance' [2]. Additionally, Imms et al. described part of the concept of participation as 'attendance', which can be regarded as the diversity of activities in which an individual takes part.[3]

In individuals with CP, participation attendance generally stabilizes whilst growing into adulthood.[4] However, participation difficulties and need for assistance tend to increase for most participation domains when individuals with CP reach their mid- and late-twenties. Particularly for participation in domestic life and interpersonal relationships, difficulties and need for assistance increase from the age of 16 years, even among those with higher gross motor capacity.[5] Despite the distinct types of environment where participation in domestic life and interpersonal relationships can take place, adults with CP are restricted in both participation domains. Compared to their peers, adults with CP show difficulties with household tasks , and a lower level of participation in romantic relationships.[5,6] This illustrates that adults with CP may grow into deficit, particularly in relation to domestic life and interpersonal relationships. Knowledge on early predictors of adult participation may help rehabilitation professionals and individuals with CP to avoid this phenomenon.

Our previous multicenter longitudinal research showed that, in addition to intellectual disability and level of gross motor function, special education, epilepsy and speech impairment are longitudinally associated with the development of participation attendance in individuals with CP from childhood into adulthood.[4,7,8] However, because these studies mainly focused on the course of development, only a few factors (mainly factors reflecting the severity of CP) were examined for their longitudinal association with participation. Cross-sectional research and a study with a 5-year follow-up in various age groups showed that it is also important to assess environmental and personal factors.[6,9-11] In children with CP, the physical home environment and the attitude of peers were strongly related to (amongst others) difficulty of participation in domestic life and interpersonal relationships, respectively.[10] A longitudinal study found that childhood psychological problems and parental stress predicted reduced participation in interpersonal relationships as adolescents with CP.[11] In addition, qualitative research among adolescents with CP indicated that self-confidence and other people's attitudes are important barriers and facilitators for social participation.[9] In young adults with CP, higher self-esteem, higher self-efficacy and a mainstream school environment were positively associated with having a romantic relationship.[6] No long-term studies have reported which teenage factors (across all components of the ICF) within an individual predict adult participation.

Therefore, the present study examines which teenage factors predict participation attendance, as well as difficulty and need for assistance with participation in domestic life and interpersonal relationships, among adults with CP without intellectual disability.

METHODS

Design

In this prospective cohort study a 13-year follow-up measurement was performed in one of the PERRIN (Pediatric Rehabilitation Research In the Netherlands) cohorts. The baseline age of the cohort was 16-20 years and previously 2- and 4-year follow-up measurements had been done. Participants had a clinical diagnosis of CP, were born in 1981-1986, and were able to participate in Dutch interviews. Exclusion criteria were having an intellectual disability, and any additional disorders affecting motor functioning. The recruitment process of the initial cohort (n=103) is reported elsewhere.[12]

Participant recruitment

Of the 103 individuals belonging to the cohort, nine had previously indicated that they no longer wished to be approached, therefore 94 individuals with CP were invited to participate in the 13-year follow-up measurement (when they were aged 28-34 years). Recruitment took place via two mailings carried out by the research team and, in case of no response, a telephone call on behalf of their treating rehabilitation clinician, or (in some cases) an additional mailing by their treating rehabilitation clinician. Finally, 54 individuals with CP signed an informed consent and participated in this 13-year follow-up study, which was approved by the Medical Ethical committee of the Erasmus University Medical Center.

Procedure

Participants were interviewed at home (or another location they selected) about their participation in domestic life and interpersonal relationships. Additionally, participants completed online questions about their age, gender, housing situation (with parents, alone, with partner), housing type (regular, adapted, assisted), romantic relationship (ever, never), children (yes, no). Baseline characteristics covering all ICF components (56 factors) were collected with various questionnaires (described in section 2.4) and included as candidate predictor variables.

Materials and instruments

Adult participation in domestic life and interpersonal relationships

Adult participation appraised the ICF domains 'domestic life' and 'particular interpersonal relationships'. The concepts assessed were participation attendance using the Vineland Adaptive Behavior Scales Second Edition survey version (Vine-II).[13] and difficulty and need for assistance with participation using the Short version of the Assessment of Life Habits (Life-H) questionnaire 3.1.[14]

Participation attendance measured with the Vine-II

To describe participation attendance in domestic life and particular interpersonal relationships we used the subdomains 'Domestic daily living skills' (21 items) and 'Interpersonal relationships' (38 items) of the Vine-II. The Vine-II measures the performance of a range of activities in these subdomains on a three-point scale: 0 'never performed', 1 'sometimes or partially performed', 2 'usually or habitually performed'. A sum score was calculated for each subdomain. Problems in participation attendance were defined as scores > 1 standard deviation (SD) lower than the mean score in the typically developing age group aged 30-39 years (score <47 for domestic living skills, and <74 for interpersonal relationships).[13] Good psychometric properties are reported for the Vine-II. [13]

Difficulty and need for assistance with participation measured with the Life-H

To describe difficulty and need for assistance with participation in domestic life and interpersonal relationships the Life-H domains ‘Housing’ (8 items) and ‘Interpersonal Relationships’ (7 items) were used. Each item consists of two parts: the degree of experienced difficulty (‘no difficulty’, ‘some difficulty’, ‘accomplished by a proxy’ or ‘not accomplished’) and the needed assistance (‘no assistance, ‘use of assistive device’, ‘adaptation’ and/or ‘with human assistance’). The two scores for each item are combined, resulting in a 10-point ordinal scale that ranges from ‘not accomplished’ to ‘no difficulty and no needed assistance’.[14] Mean scores were calculated for each domain. For both housing and interpersonal relationships a score < 8.89 indicated participation with difficulties. Good psychometric properties are reported for the Life-H.[15-18]

Candidate predictor variables

Candidate predictor variables included the earliest available assessment of the factors (i.e. preferably baseline, otherwise the assessment at 2 or 4 year follow-up). These factors were clustered according the ICF components in health condition (further referred to as ‘CP-related factors’, n=3), body functions (n=3), motor capacity (n=1), environmental factors (n=10), personal factors (n=21), and activity and participation (n=20). Categorical factors were only included if the sample assessed at the 13-year follow-up had at least two participants in each category (after dichotomizing).

CP-related factors

CP-related factors addressed the functional severity of CP using the five-level Gross Motor Function Classification System (GMFCS)[19-21] and the five-level Manual Ability Classification System (MACS),[22,23] and the type of CP (spastic *versus* dyskinesia, ataxia, and mixed). The GMFCS and MACS classification were subdivided into three levels: GMFCS level I-II (walking independently) *versus* III-IV (walking with aids or human assistance) and V (not walking); and the MACS level I-II (handling objects independently) *versus* III-IV (handling objects with help) and V (not able to handle objects).

Body functions

Body functions included educational level as a gross indication for cognitive function, vision impairment (yes *versus* no), and speech problems (no problems with speech and audible *versus* little problems but audible to not audible). Educational level was categorized as low (non-theoretical programme of pre-vocational secondary education or lower) *versus* medium-high (theoretical programme of pre-vocational secondary education, senior general secondary education, pre-university education, higher professional education and university education).

Baseline motor capacity

Motor capacity was assessed using the baseline score of the 66-item Gross-Motor Function Measure (GMFM-66).[24] Each item was scored on a four-point scale (0-3), and an interval sum score (0-100) was calculated with the GMAE software.

Environmental factors

Environmental factors included housing type (adapted *versus* non-adapted), educational and employment type, the level of parental education (as an indication for socio-economic status), parenting style, and social support. Educational and employment type was categorized as mainstream (regular primary, regular secondary education, (un)paid job) *versus* special education and employment (daycare, special primary/secondary education, no education, no (un)paid job). The level of parental education was categorized in low (no education or primary education), medium (secondary education, including senior vocational education), and high (higher education and university) and dichotomized as low-medium *versus* high. Parenting style was assessed with the short form of the EMBU (‘*Egna Minnen av Barndoms Uppfostran*’, translated as ‘My memories of upbringing’ [s-EMBU]).[25] s-EMBU assesses perceived parental rearing behavior of both parents using three scales ‘Rejection’, ‘Emotional Warmth’ and ‘Protection’. Social support was determined with the 12-item Social Support List 12-Interaction (SSL-12), which includes the domains ‘Daily support’, ‘Support problems’, ‘Appreciation’, and ‘Negative interactions’.[26] For both the s-EMBU and the SSL the extent to which statements (e.g. “My parents tried to encourage me to become the best”) were applicable was reported on a four-point scale and sum scores were calculated for each domain.

Personal factors

The personal factors included gender (male *versus* female), nationality (Dutch *versus* other), personality, perceived competence, self-efficacy, and coping style. To determine personality, participants completed the 133-item Dutch Personality questionnaire (NPV), which is derived from the California Psychological Inventory.[27,28] Items contain statements (e.g. “I trust people only when I know them well”) covering seven subscales: ‘Inadequacy’, ‘Social inadequacy’, ‘Rigidity’, ‘Hostility’, ‘Egoism’, ‘Dominance’, and ‘Self-esteem’. A sum score was calculated for each subscale. Perceived competence was assessed with scales derived from the Dutch version of Harter’s Social Perception Profile for Adolescents scale (SPPA).[29,30] The Dutch SPPA is a 35-item instrument covering the subscales: ‘Scholastic competence’, ‘Social acceptance’, ‘Athletic competence’, ‘Physical appearance’, ‘Behavioral conduct’, ‘Close friendship’ and ‘Global self-worth’. A sum score was calculated for each subscale. To determine self-efficacy, two questionnaires were used: i) the 25-item Adolescent Social Self-efficacy scale (S-EFF) that describes commonly occurring social events which may be problematic for teenagers,[31] and ii) The 12-item General Self-Efficacy Scale (G-SES-12). Sum scores were calculated for each questionnaire. To determine the coping style of the adolescent, the 21-item Coping Inventory for Stressful Situations–Short Form (CISS-21) was used.[32] The CISS-21 assesses three coping strategies: task-oriented (e.g. ‘solve the problem’), emotion-oriented (e.g. ‘become upset’), avoidant coping (e.g. ‘visit a friend’). Participants score to what extent they use coping activities when confronted with different stressful situations. A mean score was calculated for each coping strategy.

Baseline activities and participation

To determine baseline attendance to activities and participation, the Vineland Adaptive Behavior Scales (VABS)[33,34] were used (this is an older version of the Vine-II, which was not available during the baseline assessments). All subdomains of the VABS domains ‘daily living skills’, ‘socialization’, and ‘communication’ were examined as predictor variables.

To determine the difficulty and need for assistance with activities and participation at baseline the Life-H 3.0 questionnaire[18] was used (this is an older version of the Life-H 3.1 which was not available during the baseline assessments). The Life-H 3.0 has 11 domains that cover activities of daily life and social roles (nutrition, fitness, personal care, communication, housing, mobility, responsibilities, interpersonal relationships, community life, education and employment, and recreation).

Data analysis

Statistical procedures were performed with the IBM SPSS statistics, version 22.0 (SPSS^{â} Inc, Chicago, Illinois, USA). Descriptive statistics were provided for participant characteristics at 13-year follow-up and for the candidate predictor variables at baseline. To reduce measurement error, the baseline scores of motor capacity (GMFM-66), attendance to activities and participation (subdomains of the VABS), and difficulty and need for assistance with activities and participation (subdomains of Life-H 3.0) were estimated using a linear mixed model analysis including three repeated measures (baseline, and 2 and 4-year follow-up) with age as covariate.

For each of four outcomes at 13-year follow-up (attendance in, and difficulty and need for assistance with participation in domestic life and in interpersonal relationships), stepwise multiple linear regression analyses were performed to identify significant predictors. First, baseline factors within each ICF component were entered in a multiple linear regression analysis to determine significant predictors in each ICF component. To avoid multicollinearity, a forward selection procedure ($p<0.1$) was used. Second, all identified predictors in the ICF components CP-related factors, body functions and structures, motor capacity, environmental factors, and personal factors were analysed together and with a forward selection procedure an overall prediction model was developed. For each prediction model, the explained variance (R^2) was provided.

In an additional analysis we examined if baseline activities and participation (both attendance in, and difficulty and need for assistance with) improved the prediction models to identify additional factors modifiable using rehabilitation. In line with the identification of significant predictors within each ICF component, we first identified which subdomains within each domain of the questionnaire were significant predictors. With a forward selection procedure these significant baseline participation scores were then added to the identified prediction models. The baseline participation factors that significantly ($p<0.05$) contributed to the prediction model were incorporated in the extended prediction model.

RESULTS

Participants

Fifty-four adults with CP (57% of those approached) aged 29-34 years agreed to participate in the 13-year follow-up. Only one was classified as GMFCS level V and MACS V and was therefore excluded from the analysis. The characteristics of the remaining 53 persons in GMFCS level I-IV are presented in Table 1. High average scores were observed for participation in domestic life [Vine-II: 42.6 (SD 10.3) out of 48 points; Life-Habits: 8.3 (SD 1.4) out of 10 points] and for interpersonal relationships [Vine-II: 73.3 (SD 4.0) out of 76 points; Life-Habits: 9.2 (SD 1.2) out of 10 points]. Nevertheless, a large percentage of adults scored below the reference norms, ranging from 22.6% to 58.5% for difficulty and need for assistance of participation in interpersonal relationships and domestic life, respectively.

Candidate predictor variables

Table 2 presents descriptives and psychometric properties of the candidate predictor variables and the contribution of the identified significant factors for each ICF component and each outcome of participation.

Table 1: Characteristics of the participants at 13-year follow-up.

	total *N*: 53
Age in years: mean (SD); min-max	31.7 (1.4); 29.1 to 34.2
Gender (male/female)	28/25
GMFCS	38
I	4
II	4
III	7
IV	1 excluded
V	
MACS	41
I	10
II	1
III	1
IV	1 excluded
V	
Type of CP (spastic/other)	45/8
Housing situation	2
With parents	29
Alone	22
With partner	
Housing type	39
Regular housing	7
Adapted housing	6
Assisted housing[1]	1
Missing	
Relationship (ever/never/missing)	39/11/3
Children (yes/no)	12/41
Participation in Domestic life	**Mean (SD); min-max; [below reference norm][2]**
Vine-II domestic daily living skills (range 0-48)	42.6 (10.3); 4-48; [39.6%]
Life-Habits 3.1 housing (range 1-10)	8.3 (1.4); 5-10; [58.5%]
Participation in Interpersonal relationships	
Vine-II interpersonal relationships (range 0-76)	73.3 (4.0); 59-76; [35.8%]
Life-Habits 3.1 interpersonal relationships (range 1-10)	9.2 (1.2); 4-10; [22.6%]

[1] Assisted housing means that human assistance is available in the adults' house during day and/or night [2]Reference norm: Vine-II domestic living skills <47, and interpersonal relationships <74 (= - 1 SD for typically developing age group 30-39 years); Life-Habits housing and interpersonal relationships <8.89 (= with difficulties).

The MACS and nationality were excluded as candidate variables from the multiple linear regression analysis, since only one person was left in each category when combined with other factors. Receptive communication measured with the VABS was also excluded as a candidate predictor variable, since all participants had a maximum score at baseline. For the separate ICF components, the R^2 for predictors ranged from 5.4% (performance of socialization) to 62.0% (environmental factors).

Prediction models: domestic life

Table 3 presents the prediction models for attendance in, and difficulty and need for assistance with adult domestic life. Low attendance in domestic life in adulthood was predicted by lower gross motor capacity and a more protective parenting style as a teenager (R^2=60.9%). Extending the prediction model with baseline participation revealed that attendance in written communication was the strongest factor, improving R^2 with 2.3%: the higher the written communication attendance as a teenager, the higher the attendance in participation in domestic life as an adult.

More difficulty and need for assistance with domestic life in adulthood was predicted by lower gross motor capacity, a rigid personality, and special education as a teenager (R^2=64.6%). The significant baseline activity and participation factors did not improve this model.

Prediction models: interpersonal relationships

Table 4 presents the prediction models for attendance in, and difficulty and need for assistance with adult interpersonal relationships. Low attendance in interpersonal relationships in adulthood was predicted by a rejective parenting style and less daily support as a teenager (R^2=40.6%). Extending the prediction model with baseline participation, showed that attendance in personal daily living skills was the strongest factor, improving R^2 with 13.7%: The higher the attendance in personal daily living skills as a teenager, the higher the adult attendance in interpersonal relationships. With this factor included in the model the factor daily support became not significant.

More difficulty and need for assistance with interpersonal relationships in adulthood was predicted by being male, social inadequacy (i.e. a socially avoidant personality), an avoiding coping style, and a rejective parenting style as a teenager (R^2=49.6%). The significant baseline participation factors did not improve this model.

DISCUSSION

Environmental and personal factors, along with gross motor capacity (measured with the GMFM, and only for domestic life) and teenage participation in communication and daily living skills predicted participation in domestic life and/or interpersonal relationships of adults with CP. Level of functional severity, type of CP or factors concerning body functions were no significant predictors.

Comparison of predictors between domestic life and interpersonal relationships

For future participation in both domestic life and in interpersonal relationships, parenting style (protective or rejective) and personality traits (rigid or socially avoidant) were significant predictors. In addition, we found that the attending a protected school predicted more difficulty in adult participation in domestic life. This may be explained by the more protective environment that special education provides, and therefore seems to be in line with a protective parenting style predicting poorer participation in interpersonal relationships. Likewise, another study demonstrated the predictive value of childhood psychological problems.[11]

Table 2: Descriptives of the candidate predictor variables at baseline (N=53[1]), and the identified significant predictor variables for each ICF component.

			Domestic life		Interpersonal relationships	
	Psychometric properties (reliability and validity)		*Vine-II* Unstandardized β (SE), R^2%	*Life-H* Unstandardized β (SE), R^2%	*Vine-II* Unstandardized β (SE), R^2%	*Life-H* Unstandardized β (SE), R^2%
CP-related factors (health condition)		***n***				
Functional severity,	Both good (Jahnsen, et al., 2006; Palisano, et al., 1997; Wood & Rosenbaum, 2000)		GMFCS I-II=reference	GMFCS I-II=reference	GMFCS I-II=reference	GMFCS I-II=reference
GMFCS level (I + II / III + IV)		42/11	GMFCS III-IV -17.3 (2.57)$^{\#}$	GMFCS III-IV -2.17 (0.39)$^{\#}$	GMFCS III-IV -2.93 (1.30)*	GMFCS III-IV -0.81 (0.41)*
Functional severity, MACS level (I + II / III + IV); *Excluded as candidate variable*	Both good (Eliasson, et al., 2006)	51/2				
Type of CP (Spastic/Other)		45/8	R^2=47.1%	R^2=38.0%	R^2=9.1%	R^2=7.2%
Body functions and structures		***n***				
Educational level (Low/medium-high)		13/38 Missing: 2	Educational level, 6.03 (3.23)*	None	Educational level, 2.35 (1.26)*	None
Vision impairment (no/yes)		45/4 Missing: 4				
Speech (No problems with speech and audible/little problems but audible till not audible)		47/3 Missing: 3	R^2 = 6.6%		R^2 = 6.7%	
Motor capacity		**Mean (SD); min-max**				
Gross Motor Function Measure-66 (GMFM-66) (66 items, range 0-100)	Both good (Russell, et al., 2000)	82.4 (18.8); 25.7 to 100	GMFM, 0.38 (0.06)$^{\#}$	GMFM, 0.05 (0.01)$^{\#}$	None	None
			R^2 = 47.6%	R^2 = 47.3%		
Environmental factors		***n***				
Housing type (Non-adapted/adapted home)		34/10 Missing: 9	Housing type, -13.0 (2.36)$^{\#}$	Housing type, -0.82 (0.40)*	SES, 1.69 (0.94)**	Rejection, -3.96 (0.79)$^{\#}$
Educational type (Mainstream/special education)		32/12 Missing: 9	Protection, -0.83 (0.23)$^{\#}$	Educational type, -1.87 (0.38)$^{\#}$	Rejection, -0.73 (0.27)*	Appreciation, 0.15 (0.09)*
Level of parental education (Low-medium/high)		32/19 Missing: 2			Daily support, 0.74 (0.26)$^{\#}$	

Table 2: Continued

			Domestic life		Interpersonal relationships	
	Psychometric properties (reliability and validity)		*Vine-II*	*Life-H*	*Vine-II*	*Life-H*
			Unstandardized β (SE), R^2%	Unstandardized β (SE), R^2%	Unstandardized β (SE), R^2%	Unstandardized β (SE), R^2%
Environmental factors		**Mean (SD); min-max**				
Parenting style (s-EMBU) [2,5]	Both good (Aluja, et al., 2006)					
- Rejection (7 items, range 7-28)		8.4 (2.3); 6.0 to 16.0	R^2 = 62.0%	R^2 = 51.4%	R^2 = 36.6%	R^2 = 43.5%
- Emotional warmth (6 items, range 6-24)		19.0 (3.2); 11.0 to 24.0				
- Protection (10 items, range 10-40)		20.5 (4.2); 14.0 to 38.0				
		Missing: 4				
Social support (Social Support List 12-Interaction (SSL-12)) [3]	Good reliability (Van Sonderen, 1993), no information on validity					
- Daily support (4 items, range 4-16)		11.8 (1.8); 7.0 to 16.0				
- Support problems (4 items, range 4-16)		11.4 (2.1); 7.0 to 16.0				
- Appreciation (4 items, range 4-16)		11.4 (1.8); 8.0 to 16.0				
- Negative interactions (7 items, range 7-28)		11.0 (2.8); 7.0 to 19.0				
		Missing: 8				
Personal factors		***n***				
Gender (male/female)		28/25	None	Rigidity, -0.06 (0.02)**	Gender, 3.03 (0.79)$^{\#}$	Gender, 0.80 (0.26)$^{\#}$
Nationality (Dutch/other) *Excluded as candidate variable*		42/2 Missing: 9		Self-esteem, 0.09 (0.03)$^{\#}$	Social inadequacy, -0.21 (0.05)$^{\#}$	Social inadequacy, -0.08 (0.02)$^{\#}$
Personality, Dutch Personality questionnaire (NPV) [2]	Both good (Gough, 1996; Luteijn F, 2001)	**Mean (SD); min-max**			Scholastic competence, 0.28 (0.14)**	Avoidance coping, -0.52 (0.22)**
- Inadequacy (21 items, range 0-42)		11.9 (7.6); 0 to 30			Behavioral conduct, -0.43 (0.16)**	
- Social inadequacy (15 items, range 0-30)		9.3 (7.3); 0 to 28				

Table 2: Continued

			Domestic life		**Interpersonal relationships**	
	Psychometric properties (reliability and validity)		*Vine-II* Unstandardized β (SE), R^2%	*Life-H* Unstandardized β (SE), R^2%	*Vine-II* Unstandardized β (SE), R^2%	*Life-H* Unstandardized β (SE), R^2%
Personal factors		**Mean (SD); min-max**				
- Rigidity (25 items, range 0-50)		26.9 (7.6); 9 to 41		$R^2 = 40.6\%$	$R^2 = 46.3\%$	$R^2 = 37.4\%$
- Hostility (19 items, range 0-38)		15.0 (6.2); 2 to 32				
- Egoism (16 items, range 0-32)		10.6 (4.4); 1 to 20				
- Dominance (17 items, range 0-34)		16.1 (5.4); 3 to 25				
- Self-esteem (19 items, range 0-38)		28.0 (6.6); 9 to 38				
		Missing: 4				
Perceived competence, Harter's Social Perception Profile for Adolescents scale (SPPA)	Both good (Treffers, et al., 2002)					
- Scholastic competence (5 items, range 5-20)		14.3 (2.9); 7 to 20				
- Social acceptance (5 items, range 5-20)		14.2 (3.0); 7 to 20				
- Athletic competence (5 items, range 5-20)		10.2 (3.2); 5 to 18				
- Physical appearance (5 items, range 5-20)		13.2 (3.6); 5 to 19				
- Behavioral conduct (5 items, range 5-20)		16.5 (2.7); 10 to 20				
- Close friendship (5 items, range 5-20)		17.1 (3.0); 9 to 20				
- Global self-worth (5 items, range 5-20)		15.3 (3.2); 5 to 20				
		Missing: 1				
Self-efficacy, General Self-Efficacy Scale (G-SES-12) [3] (12 items, range 12-60)	No information on reliability and validity	45.5 (6.2); 32 to 56 Missing: 6				
Social self-efficacy, Adolescent Social Self-efficacy scale (S-EFF) [2] (25 items, range 25-175)	No information on reliability and validity	135.0 (24.1); 78 to 175 Missing: 4				

Table 2: Continued

	Psychometric properties (reliability and validity)		Domestic life		Interpersonal relationships	
			Vine-II Unstandardized β (SE), R²%	*Life-H* Unstandardized β (SE), R²%	*Vine-II* Unstandardized β (SE), R²%	*Life-H* Unstandardized β (SE), R²%
Personal factors		**Mean (SD); min-max**				
Coping style, Coping Inventory for Stressful Situations (CISS-21)	No information on reliability and validity, good internal consistency (Cohan, Jang, & Stein, 2006; N. S. Endler, Speer, Johnson, & Flett, 2000)					
- Task oriented (7 items, range 1-7)		3.6 (0.7); 2.0 to 5.0				
- Emotion oriented (7 items, range 1-7)		2.8 (0.7); 1.6 to 5.0				
- Avoidance (7 items, range 1-7)		3.1 (0.6); 1.4 to 4.4 Missing: 1				
Activities and participation as teenager		**Mean (SD); min-max**				
VABS Communication	Both good (De Bildt, 2003; S. S. Sparrow, et al., 1984)		Written, 2.41 (1.03)**	Written, 0.29 (0.14)**	Written, 0.98 (0.37) **	Expressive, 1.56 (0.78)*
- Receptive *(13 items, range 0-26)* [4]		26.0 (0.0); 26.0 to 26.0				
- Expressive *(31 items, range 0-62)*		61.73 (0.21); 60.93 to 61.89	R² = 9.8%	R² = 7.5%	R² = 12.2%	R² = 7.2%
- Written *(23 items, range: 0-46)*		43.87 (1.34); 39.63 to 45.36				
VABS Daily living skills	Both good (De Bildt, 2003; S. S. Sparrow, et al., 1984)		Domestic, 1.13 (0.22)#	Personal, 0.49 (0.24)**	Personal, 1.97 (0.50)#	Community, 0.08 (0.04)*
- Personal *(39 items, range 0-78)*		77.15 (0.92); 73.71 to 77.78		Domestic, 0.12 (0.04)**		
- Domestic *(21 items, range 0-42)*		31.92 (4.57); 18.23 to 39.44				
- Community *(32 items, range 0-64)*		51.73 (3.77); 45.24 to 56.95	R² = 34.6%	R² = 40.9%	R² = 23.6%	R² = 5.7%
VABS Socialization	Both good (De Bildt, 2003; S. S. Sparrow, et al., 1984)		Play and leisure, 1.35 (0.79)*	Interpersonal relationships, 0.34 (0.16)**	Interpersonal relationships, 1.68 (0.39)#	Interpersonal relationships, 0.25 (0.13)*
- Interpersonal relationships *(28 items, range: 0-56)*		51.15 (1.21); 48.32 to 53.75				

Table 2: Continued

	Psychometric properties (reliability and validity)		**Domestic life**		**Interpersonal relationships**	
			Vine-II Unstandardized β (SE), R^2%	*Life-H* Unstandardized β (SE), R^2%	*Vine-II* Unstandardized β (SE), R^2%	*Life-H* Unstandardized β (SE), R^2%
Activities and participation as teenager		**Mean (SD); min-max**				
- Play and leisure *(20 items, range: 0-40)*		37.81 (1.78); 29.07 to 39.25	$R^2 = 5.4\%$	$R^2 = 8.3\%$	$R^2 = 26.4\%$	$R^2 = 7.4\%$
- Coping skills (*18 items, range: 0-36)*		35.46 (0.86); 31.60 to 36.40				
Life-H 3.0 Activities	Both good (Desrosiers, et al., 2004; Fougeyrollas, et al., 1998; Lemmens, et al., 2007; Magasi & Post, 2010)		Mobility, 5.32 (0.64)$^{\#}$	Mobility, 0.72 (0.09)$^{\#}$	Nutrition, 1.90 (0.65)$^{\#}$	Communication, 2.53 (0.53)$^{\#}$
- Nutrition *(3 items, range: 0-10)*		9.03 (1.26); 4.56 to 9.75			Fitness, 1.45 (0.45)*	
- Fitness *(3 items, range: 0-10)*		9.26 (0.77); 5.14 to 9.79			Personal care, -1.16 (0.45)**	
- Personal care *(7 items, range: 0-10)*		9.06 (1.93); 1.84 to 9.96			Communication, 5.32 (1.89)$^{\#}$	
- Communication *(7 items, range: 0-10)*		9.84 (0.27); 8.59 to 9.97				
- Housing *(8 items, range: 0-10)*		9.38 (0.96); 5.66 to 9.93	$R^2 = 57.4\%$	$R^2 = 54.9\%$	$R^2 = 44.0\%$	$R^2 = 30.9\%$
- Mobility *(5 items, range: 0-10)*		8.68 (1.47); 3.61 to 9.85				
Life-H 3.0 Social roles	Both good (Desrosiers, et al., 2004; Fougeyrollas, et al., 1998; Lemmens, et al., 2007; Magasi & Post, 2010)		Recreation, 3.29 (0.49)$^{\#}$	Education and Employment, 0.85 (0.43)*	Interpersonal relationships, 2.08 (0.77)**	Interpersonal relationships, 0.87 (0.24)$^{\#}$
- Responsibilities *(6 items, range: 0-10)*		9.55 (0.61); 7.05 to 9.87		Recreation, 0.39 (0.08)$^{\#}$	Community life, 0.98 (0.38)**	
- Interpersonal relationships *(7 items, range: 0-10)*		9.58 (0.65); 5.78 to 9.92				
- Community life *(6 items, range: 0-10)*		8.79 (1.34); 4.54 to 9.71	$R^2 = 46.6\%$	$R^2 = 52.7\%$	$R^2 = 30.9\%$	$R^2 = 21.2\%$
- Education and Employment *(10 items, range: 0-10)*		8.67 (0.39); 7.52 to 9.21				
- Recreation *(6 items, range: 0-10)*		8.69 (2.15); 0.91 to 9.97				

[1] N=53, since the one participant classified as GMFCS level V was excluded from the analysis; [2] Assessed at 2-year follow-up, age 18-22 years; [3] Assessed at 4-year follow-up, age 20-26 years; [4] Raw data, as modelling was not possible. [5] Egna Minnen Barndoms Uppfostran (English: My memories of upbringing); $^{\#}$ <0.01, ** = <0.05, *=<0.1

Additionally, in a qualitative study, youth with CP indicated supporting parents and a flexible, sociable personality were enhancing, while strict parents or professionals and avoidant coping were limiting their participation experiences.[37] Assessing a person's personality traits and coping style may help identify those teenagers with CP at risk for future suboptimal participation. Furthermore, it is known from the general population that the development of personality of typically developing children into adolescence benefits from positive interactions between mother and child.[38] This underlines the importance of an encouraging parenting style of parents of persons with CP, which may give teenagers the freedom to explore opportunities for optimal participation in domestic life and interpersonal relationships. This recommendation is not restricted to parents of teenagers only, since the used questionnaire reflects memories of one's upbringing in general and parenting starts at a very young age.[39]

Gross motor capacity (measured with the GMFM-66) predicted domestic life but not interpersonal relationships, whereas daily support and coping style only predicted interpersonal relationships. This can be explained by the nature of the domains: domestic life includes mobility-related situations (e.g. cleaning the room), whereas interpersonal relationships reflect life situations in which persons have to react to or anticipate others. Similar to our results, the SPARCLE study also showed that impairment (including walking ability) predicted a large part of domestic life in and the attitude of others was strongly associated with difficulty in interpersonal relationships in adolescents with CP.[10,11] The present results show that the different domains of participation (domestic life and interpersonal relationships) have their own domain-specific predictors besides the more common predictors of parenting style and personality. Depending on the preferred participation domain to target, teenager rehabilitation may emphasize improving gross motor capacity and/or learning to cope with other people's attitudes.

Extending models with baseline activities and participation

Similar to Dang et al., adding baseline activity and participation attendance decreased regression coefficients of environmental and personal factors.[11] It should be noted that high univariate associations with future participation, and multicollinearity between several participation domains at baseline (data not presented) do not allow us to conclude that written communication and self-care skills are the only relevant domains for future participation. Rather, the level of activities and participation as a teenager in a broad sense seems important for future participation as an adult, both for attendance and for difficulties in participation. Thus, to optimize adult participation, rehabilitation professionals may focus on improving activities and participation of teenagers with CP. This may involve discussing with teenagers which participation areas are relevant for them to establish their personal goals and addressing individual challenges.

Strengths and limitations

To our knowledge this is the first study to examine teenage predictors of participation of adults with CP in a long-term follow-up. The study design allowed an investigation of a large number of candidate predictor variables across all ICF components, resulting in teenage predictors that explain a high percentage of the variance of adult participation. The relatively homogeneous sample of individuals without intellectual disability, with many adults classified as GMFCS level I, allowed us to explore which other types of factors, besides functional severity, predict participation. Nevertheless, a limitation of the study is the relatively small sample size, taking this into account, one should be careful generalizing results. Our results are restricted to objective qualifiers of participation (i.e. attendance to, and difficulty and need for assistance). To complement

Table 3: Prediction models for domestic life measured with the Vine-II (n=50) and the Life-H (n=44).

	Domestic life Attendance (Vine-II)				**Domestic life Difficulty (Life-H)**			
	Unstandardized Beta (SE)	Standardized Beta	p-value	R^2	Unstandardized Beta (SE)	Standardized Beta	p-value	R^2
				60.9%				64.6%
Gross motor capacity (GMFM-66)	0.30 (0.05)	0.61	<0.001		0.03 (0.01)	0.42	0.003	
Educational type (mainstream/special)	-	-	-		-1.06 (0.41)	-0.34	0.015	
Protective parenting style (s-EMBU)	-0.77 (0.20)	-0.36	<0.001		-	-	-	
Rigidity (personality; NPV)	-	-	-		-0.06 (0.02)	-0.33	0.002	
Extended prediction model (n=50)				63.2%				
Gross motor capacity (GMFM-66)	0.29 (0.05)	0.58	<0.001		-	-	-	
Protective parenting style (s-EMBU)	-0.64 (0.22)	-0.29	0.005		-	-	-	
Rigidity (NPV)	-	-	-		-	-	-	
VABS written communication as teenager	1.48 (0.71)	0.21	0.042		-	-	-	

Table 4: Prediction models for interpersonal relationships measured with the Vine-II (n=46) and the Life-H (n=50)

	Interpersonal relationships Attendance (Vine-II)				**Interpersonal relationships Difficulty (Life-H)**			
	Unstandardized Beta (SE)	Standardized Beta	p-value	R^2	Unstandardized Beta (SE)	Standardized Beta	p-value	R^2
				40.6%				49.6%
Rejection (s-EMBU)	-0.86 (0.26)	-0.43	0.002		-0.18 (0.06)	-0.36	0.002	
Daily support (SSL-12)	0.76 (0.27)	0.36	0.008		-	-	-	
Gender	-	-	-		0.71 (0.24)	0.33	0.005	
Social inadequacy (NPV)	-	-	-		-0.07 (0.02)	-0.48	<0.001	
CISS-21 Avoidance coping	-	-	-		-0.52 (0.20)	-0.30	0.013	
Extended prediction model (n=46)				54.3%				
Rejection (s-EMBU)	-0.91 (0.23)	-0.45	<0.001		-	-	-	
Daily support (SSL-12)	-0.39 (0.24)	0.20	0.104		-	-	-	
VABS personal daily living skills as teenager	2.49 (0.51)	0.51	<0.001		-	-	-	

our knowledge on this important outcome, future studies may investigate long-term predictors of subjectively experienced participation of adults with CP, such as feeling involved and engaged.

CONCLUSION

Environmental and personal factors, along with gross motor capacity (only for domestic life) and teenage participation were identified as predictors of participation of adults with CP. To optimize adult participation in domestic life and interpersonal relationships, rehabilitation professionals may support an encouraging parenting style, the ability to cope with one's own personality, and focus on improving activities and participation of teenagers with CP. Additionally, optimizing gross motor capacity as a teenager seems more important for domestic life than for interpersonal relationships.

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9

General discussion

The goal of rehabilitation is to improve functioning of individuals with disabilities in daily life. Recently, research interest for functioning and health of adults with cerebral palsy (CP) has increased. In line with this background, this thesis focused on health issues, activity and participation of young adults with CP, by performing a long-term follow-up of existing cohorts of children and teens with CP. First, we aimed to describe health issues in young adulthood. Second, we described the long-term development of activity and participation throughout young adulthood. This development was studied for several domains of activity and participation according to the ICF (International Classification of Functioning Disability and Health) and for several qualifiers of activity and participation: performance, difficulty and autonomy. Finally, we addressed early predictors of participation in young adulthood. The main findings are displayed in Figure 1 and will be discussed in this chapter.

Health issues

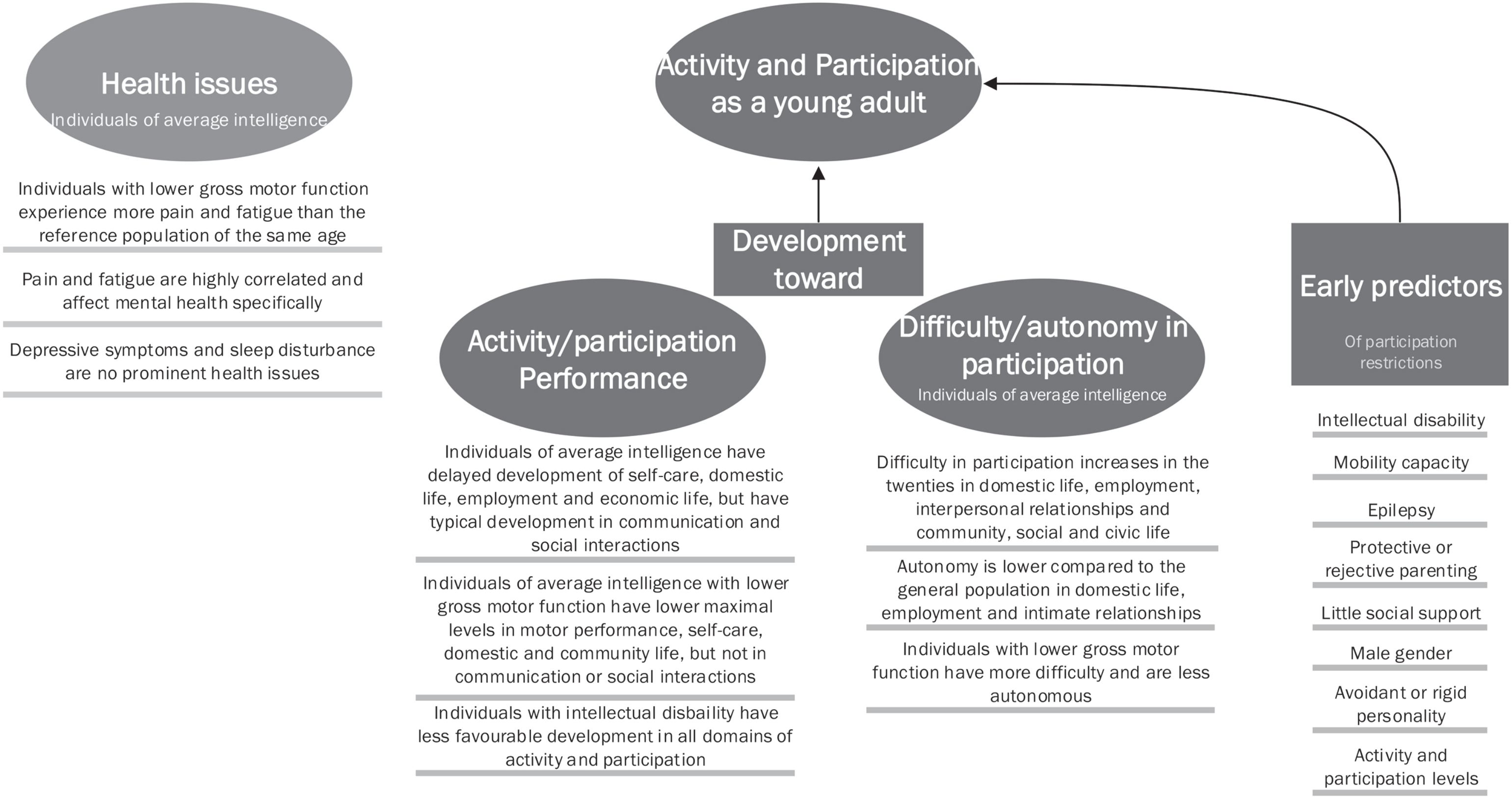
In Chapter 2 we studied pain, fatigue, depressive symptoms and sleep disturbance in individuals with CP in their twenties. Individuals with Gross Motor Function Classification System (GMFCS) level I had less pain, fatigue, depressive symptoms and sleep disturbances, while individuals with GMFCS level II-V had more pain and those with GMFCS level III-V had more severe fatigue than the age-matched general population (Figure 1, left panel). The relatively low occurrence of health issues in young adults with GMFCS level I compared to reference groups may be explained by the lifelong aspect of their disability. Individuals with CP are used to functioning with their disability, may have struggled with health issues in the past, and could therefore rate their current experienced health more neutral, in line with the mechanism of the response shift theory.[1] This theory explains that disability may lead to adjustments (recalibration or reprioritization) in internal standards or values of perceived health.[1] GMFCS subgroups were not previously compared to references, but studies similarly found more pain and fatigue in individuals with GMFCS level II-IV compared to level I. [2,3] We recommend health professionals to monitor pain and fatigue in young adults with GMFCS levels II-V. Depressive symptoms and sleep disturbance were not worse in individuals with CP compared to the general population and therefore do not seem to demand a specific approach in young adults with CP.

Pain and fatigue were more strongly inter-related in individuals with CP compared to a reference population, and also related more strongly to mental health. Health professionals may consider treating them in combination when both issues occur. A lifestyle intervention program to stimulate physical activity may for instance be adequate, since this was previously found to reduce both fatigue and pain.[4] Furthermore, future research may determine the underlying mechanism of the association between pain and fatigue. Moreover, one of our future research aims is to determine which factors, in addition to GMFCS level II-V, predict occurrence and severity of pain and fatigue in adulthood. This matches one of the aims of a recently developed patient-centered research agenda for CP, which calls for determining variables that are important to monitor or treat early on in life to prevent development of pain and fatigue later in life.[5]

Development of activity and participation into adulthood

Over the past decades, rehabilitation medicine has broadened its scope from being primarily disability-centered to evaluating an individual and its life.[6] This shift has been supported widely, and participation is now considered an important or even the ultimate health outcome by individuals with CP and rehabilitation professionals.[5,7,8] However, the definition of participation and its measurement are subject of an ongoing debate.[9-12] It is argued that although participation

Figure 1: Main results of health issues, activity and participation of young adults with cerebral palsy



Individuals with lower gross motor function experience more pain and fatigue than the reference population of the same age

Pain and fatigue are highly correlated and affect mental health specifically

Depressive symptoms and sleep disturbance are no prominent health issues

Individuals of average intelligence have delayed development of self-care, domestic life, employment and economic life, but have typical development in communication and social interactions

Individuals of average intelligence with lower gross motor function have lower maximal levels in motor performance, self-care, domestic and community life, but not in communication or social interactions

Individuals with intellectual disbaility have less favourable development in all domains of activity and participation

Difficulty in participation increases in the twenties in domestic life, employment, interpersonal relationships and community, social and civic life

Autonomy is lower compared to the general population in domestic life, employment and intimate relationships

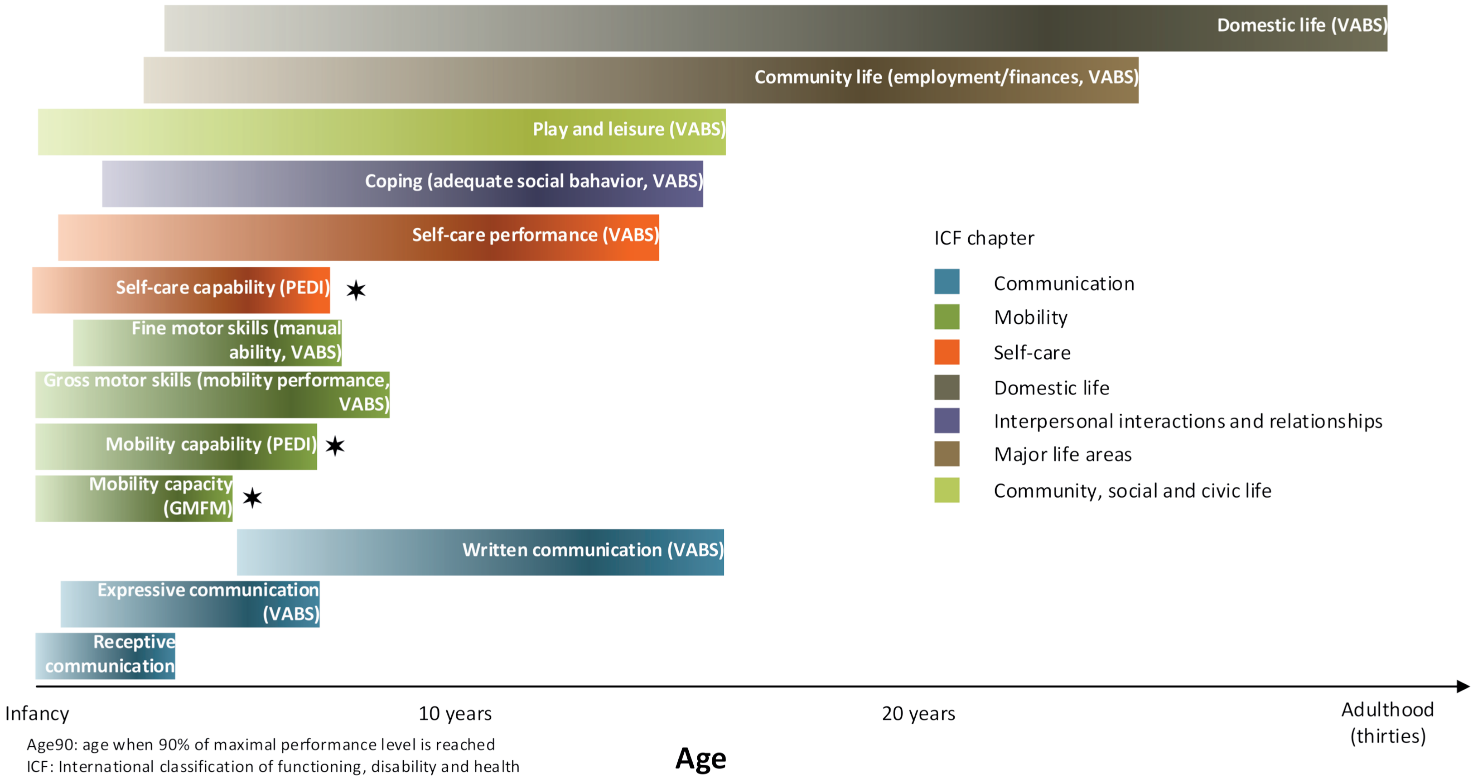
Individuals with lower gross motor function have more difficulty and are less autonomous has gained attention in research, outcome measures do not always reflect participation well.[13] Nevertheless, using the definitions and framework of the ICF, we thoroughly assessed the development and predictors of participation. We used three patient-reported outcomes to address several domains of activity and participation. The Vineland Adaptive Behavior Scales (VABS) assessed performance, registering whether or not activities in different domains of activity and participation were performed. The assessment of Life Habits (Life-H) was used to assess difficulty in participation performance. Finally, the Rotterdam Transition Profile (RTP) was used to assess autonomy in participation. The main results on development of activity and participation are displayed in the middle panel of Figure 1 and will be discussed in this section.

Performance

Chapters 3 and 4 described development of performance of activity and participation, i.e. whether or not an individual performs activities in his or her daily life, for the domains communication, mobility, self-care, domestic life, interpersonal interactions and major life areas (employment and economic life).[14] Compared to previous reports, the current study with its long-term follow-up into the early thirties allowed to more accurately estimate the limits and rate of development of individuals with CP.[15-17] Furthermore, we described development more specifically by domains of activity and participation.[15-17] We found that individuals with CP reached their maximal level of mobility performance, receptive and expressive communication in childhood. As illustrated in Figure 2a, for those of average intelligence, performance of written communication, self-care and social interactions continued to develop in adolescence, and employment, economic life and domestic activities further continued to develop in adulthood. For these individuals of average intelligence, development seemed delayed compared to reference values for mobility, self-care, domestic life, employment and economic life. For mobility, and to a lesser extent for these other domains, maximal levels of activity and participation were lower for those with lower levels of motor function. Development of communication and social interactions showed similar patterns as the general population, regardless of GMFCS level. Apparently, the disorders associated with CP affect the development of mobility, daily activities and employment, while these on average do not affect development of basic communication skills and social interactions as measured using the VABS. This contrasts a previous study that showed a strong association between gross motor function and communication skills, but this study did not correct for intellectual disability.[18] Other aspects of communication and social interactions may nevertheless be restricted in individuals with CP of average intelligence, since the VABS disregards the *quality* of communication, and individuals with CP were found to experience limitations in romantic relationships, which were not included in our results of interpersonal relationships.[19]

For individuals with CP of average intelligence, the results presented in this thesis on development of activity and participation showed that after approaching maximal levels of development in mobility capacity as measured by the gross motor function measure (GMFM) at ages 4-6 years[20] and mobility and self-care capability as measured by the pediatric evaluation of disability inventory (PEDI) at ages 5-7 years,[21] they continue to develop *performance* of activity and participation (VABS) well into adulthood. Figure 2a illustrates this pattern of development, showing the onset of development until the average age when 90% of the limit is reached for development of capacity, capability and performance of activity and of participation in several domains.[21,22] Healthcare providers should be aware that development of these individuals continues long after mobility capacity limits have been reached in performance of several domains of participation.

Figure 2a: Development from onset to Age90 for domains of activity and participation of individuals with cerebral palsy of average intelligence with GMFCS level I-IV



Age90: age when 90% of maximal performance level is reached
ICF: International classification of functioning, disability and health
GMFCS: Gross Motor Function Classification System
VABS: Vineland Adaptive Behavior Scales
PEDI: Pediatric Evaluation of Disability Inventory
GMFM: Gross Motor Function Measure
✶ : Results outside of this thesis

Individuals with intellectual disability had much less favourable development of performance in all domains except mobility, illustrated by lower maximal levels, attained at a younger age compared to individuals of average intelligence (see Figure 2a and 2b). These lower levels of performance are in line with previous studies,[15-17] but we added the finding that this development levelled off at a young age (Figure 2b) and remained relatively stable into young adulthood. Individuals with intellectual disability seem to have distinctly different needs than those of average intelligence, and the aims of rehabilitation care may be very different. Their rehabilitation treatment should be aimed at optimizing their participation and independence in daily activities by creating opportunities while offering adequate support and resources. Intellectual disability occurs frequently in individuals with CP,[23,24] but the body of literature on individuals with CP with intellectual disability is very limited. The lack of knowledge of this subgroup may be caused by methodological problems in assessment. Different methods of assessment are needed to identify treatment priorities and strategies to optimize participation in these individuals with CP with intellectual disability. Recently, assessments have been developed for physical fitness of individuals with intellectual disability,[25] but similar outcomes for activity and participation and for individuals with CP specifically are lacking. Future studies on individuals with CP with intellectual disability should carefully consider meaningful outcomes and valid ways to assess these.

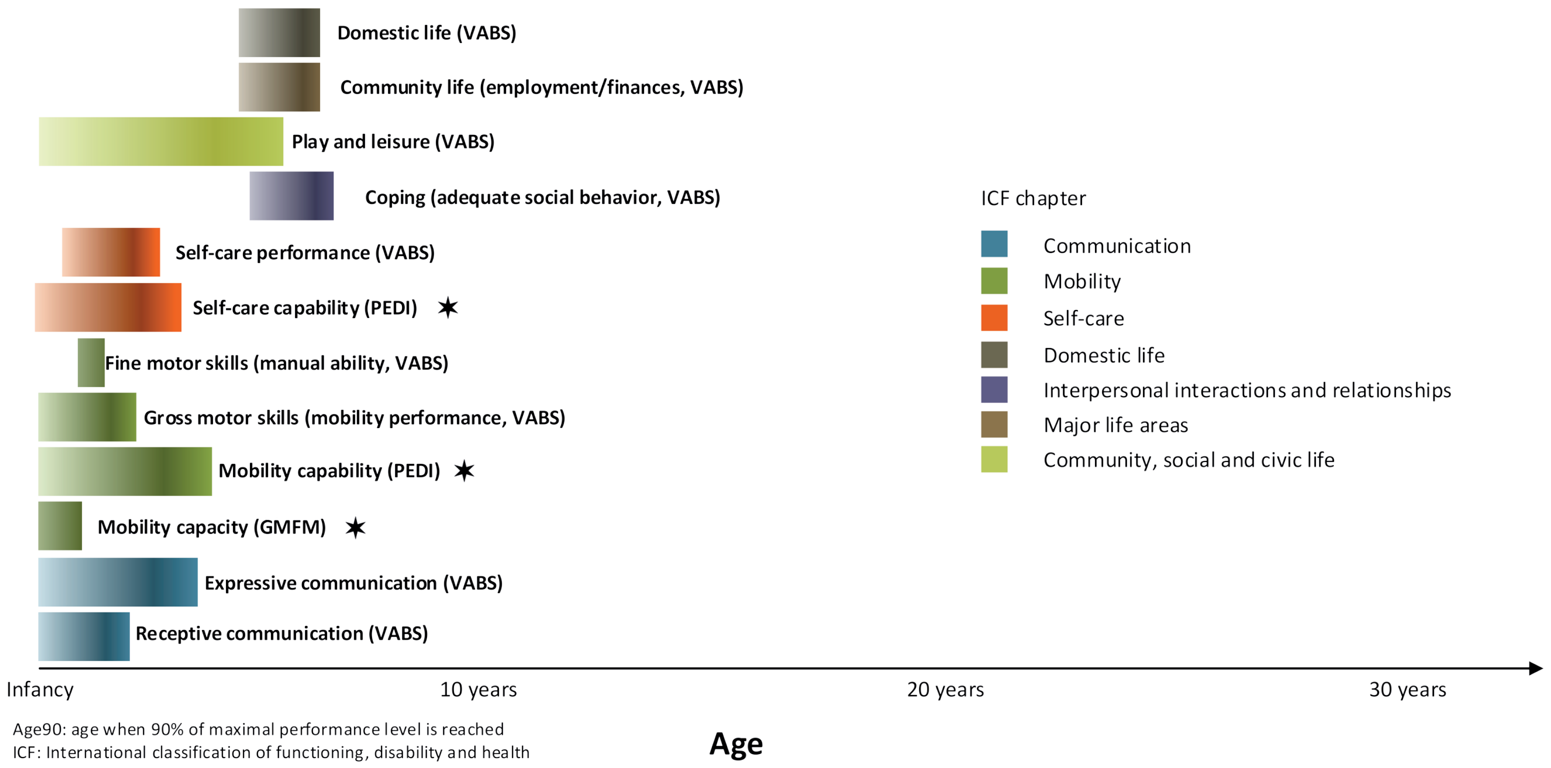
Difficulty

In Chapter 5 the longitudinal course of *difficulty* in participation of individuals with CP of average intelligence was described from adolescence into the early thirties. Those with GMFCS level IV experienced more difficulty than those with GMFCS level I regardless of age. Moreover, despite reaching high levels of performance in participation as measured by the VABS, difficulty in participation increased in the mid- and late twenties of individuals with CP of average intelligence in all domains of participation, regardless of GMFCS level. This may be induced by changes in expectations related to adult roles; desired or socially defined roles are likely to change in this phase: from being dependent of ones parents to for instance living independently, being employed and maintaining a stable relationship.[26] The increase of difficulty in the twenties may be understood using a theory that was previously suggested in the literature: growing into deficit.[8,27] This mechanism describes that problems may start to occur as an individual grows into adult roles because of age-related change of desired or socially defined roles, when individuals receive less immediate support of their parents or a teacher.

Autonomy

In addition to performance and difficulty of participation, the development of autonomy in participation for individuals with CP from youth into adulthood was described in Chapter 6. Regardless of age, individuals with GMFCS levels III-V were less often autonomous in transportation and intimate relationships compared to those with GMFCS levels I-II. In the late twenties differences between those with GMFCS levels I-II and levels III-V also emerged in employment and domestic life. The present study was the first to describe the course of autonomy for subgroups of different levels of motor function over a large age-range. Compared to age-matched references, individuals with CP were less often autonomous in their teens until their early thirties. This indicates that restrictions in independent living and intimate relationships that were previously found in a subsample of the cohort remained present in adulthood, in particular in those with GMFCS levels III-V.[27,28] Those with GMFCS level I-II seem to lag behind compared to the general population in development, but do achieve autonomy in adulthood in all domains except intimate relationships.

Figure 2b: Development from onset to Age90 for domains of activity and participation of individuals with cerebral palsy with intellectual disability and GMFCS level V



Age90: age when 90% of maximal performance level is reached
ICF: International classification of functioning, disability and health
GMFCS: Gross Motor Function Classification System
VABS: Vineland Adaptive Behavior Scales
PEDI: Pediatric Evaluation of Disability Inventory
GMFM: Gross Motor Function Measure
✶ : Results outside of this thesis

Implications and recommendations

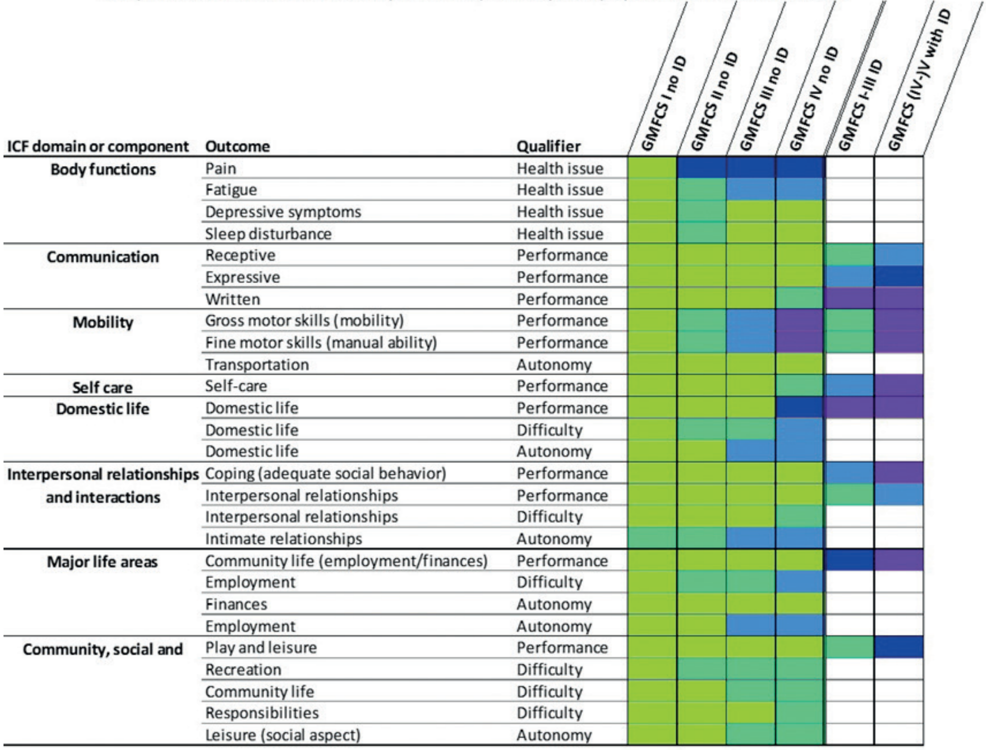
These results of ongoing development into adulthood and limitations in subgroups of young adults with CP in activity and participation support the need for adopting a lifespan perspective in the care for individuals with CP.[29,30] In current rehabilitation treatment however, a remarkable decrease in the use of rehabilitation services by individuals with CP is observed as they become adults and make the transition to adult care.[31,32] This may be due to the challenges that this health-care transition brings but also to the lack of knowledge on the care needs of adults with CP.[31] A healthcare transition period to accommodate individuals with CP from pediatric to adult care should be started early and involve all professionals, the young individual with CP and their family.[33] In addition, individuals with cerebral palsy who grow into adult roles should get specific personalized attention to support their development of participation and autonomy. Support may include transition programs in specific domains of participation such as employment and intimate relationships.[4,34,35] Some institutions in the Netherlands have already implemented specialized care for emerging adults and some recommendations are included in the rehabilitation guidelines for children with spastic CP.[36] Still, these transition programs currently lack strong evidence of effective content. Research focusing on experiences from youth and families during transition processes are called for to determine which support is wanted.[37] This information may be used to adjust the existing programs or to develop new interventions, the effect of which should be evaluated in larger effect studies. Furthermore, to stimulate active participation of adolescents and young adults in their care management, they need to be informed well of possible challenges in their participation development.

Although not described in this thesis, more subjective aspects of participation, such as participation preferences or satisfaction with their participation are additionally important to consider in clinical decision-making, especially in emerging adults.[38] Some authors argue that available studies have mainly included outcomes of objectively or quantitatively measured participation, but too often neglect subjective aspects such as a person's desire or satisfaction or the environment in which participation takes place.[12,39,40] Future studies may focus more on studying subjective experiences of participation qualitatively, and additionally aim to identify changes in the subjective aspects of participation as individuals with CP transition into adult life, so rehabilitation care may be fitted better to their age-specific needs.

Health issues and limitations in activity and participation of young adults with CP

This thesis, based on the PERRIN cohort studies, provides a broad overview of the future outlook of health issues, activity and participation of individuals with CP in their twenties. Because of a deterioration in functioning and health that was found in adults with CP of higher age,[41] these results cannot be generalized beyond this age. Considering all results from this thesis, we found that differences exist in experienced limitations as a young adult between subgroups of individuals with CP and between domains of activity and participation (Figure 3). Individuals with CP of average intelligence with GMFCS level I in their late twenties on average have few limitations; they have low levels of pain and fatigue, high levels of performance and low levels of difficulty of activity and participation in all domains. They are autonomous in transportation, domestic life, employment, finances and leisure, but less often have experience with sexual relationships than individuals in their twenties in the general population. In young adults

Figure 3: Outcomes of health issues, activities and participation of young adults with CP. Proportions of individuals without health issue, who are autonomous or the mean performance/ difficulty score proportionate to the maximal score



ICF domain or component	Outcome	Qualifier	GMFCS I no ID	GMFCS II no ID	GMFCS III no ID	GMFCS IV no ID	GMFCS I-III ID	GMFCS (IV-)V with ID
Body functions	Pain	Health issue	>90%	30-50%	30-50%	30-50%		
	Fatigue	Health issue	>90%	70-90%	50-70%	50-70%		
	Depressive symptoms	Health issue	>90%	70-90%	>90%	>90%		
	Sleep disturbance	Health issue	>90%	70-90%	>90%	>90%		
Communication	Receptive	Performance	>90%	>90%	>90%	>90%	70-90%	50-70%
	Expressive	Performance	>90%	>90%	>90%	>90%	50-70%	30-50%
	Written	Performance	>90%	>90%	>90%	70-90%	<30%	<30%
Mobility	Gross motor skills (mobility)	Performance	>90%	70-90%	50-70%	<30%	70-90%	<30%
	Fine motor skills (manual ability)	Performance	>90%	70-90%	50-70%	<30%	70-90%	<30%
	Transportation	Autonomy	>90%	>90%	>90%	>90%		
Self care	Self-care	Performance	>90%	>90%	>90%	70-90%	50-70%	<30%
Domestic life	Domestic life	Performance	>90%	>90%	>90%	30-50%	<30%	<30%
	Domestic life	Difficulty	>90%	70-90%	70-90%	50-70%		
	Domestic life	Autonomy	>90%	>90%	50-70%	50-70%		
Interpersonal relationships and interactions	Coping (adequate social behavior)	Performance	>90%	>90%	>90%	>90%	50-70%	<30%
	Interpersonal relationships	Performance	>90%	>90%	>90%	>90%	70-90%	50-70%
	Interpersonal relationships	Difficulty	>90%	>90%	>90%	70-90%		
	Intimate relationships	Autonomy	70-90%	70-90%	50-70%	50-70%		
Major life areas	Community life (employment/finances)	Performance	>90%	>90%	>90%	>90%	30-50%	<30%
	Employment	Difficulty	>90%	70-90%	70-90%	50-70%		
	Finances	Autonomy	>90%	>90%	>90%	>90%		
	Employment	Autonomy	>90%	>90%	50-70%	50-70%		
Community, social and	Play and leisure	Performance	>90%	>90%	>90%	>90%	70-90%	30-50%
	Recreation	Difficulty	>90%	70-90%	70-90%	70-90%		
	Community life	Difficulty	>90%	>90%	70-90%	70-90%		
	Responsibilities	Difficulty	>90%	>90%	>90%	70-90%		
	Leisure (social aspect)	Autonomy	>90%	>90%	70-90%	70-90%		

Legend

Colors	Measurement instruments of qualifiers
>90%	Health issue: patient-reported outcomes measurement information system (PROMIS)
70-90%	Performance: vineland adaptive behavior scales (VABS)
50-70%	Difficulty: assessment of life habits (Life-H)
30-50%	Autonomy: Rotterdam Transition Profile (RTP)
<30%	

with GMFCS levels II-V of average intelligence, more limitations occur in those with lower levels of motor function. They have higher levels of pain and fatigue, and experience difficulty and limitations in autonomy, especially in domestic life, employment and sexual relationships. Those with intellectual disability experience severe limitations in performance of activity and participation. More limitations in activity and participation in those with more severe motor function limitations and intellectual disability are in line with the literature.[42-46]

Early predictors of participation

This thesis was the first to determine predictors of participation in individuals with CP on the long-term (over 13 years) in adulthood. We studied two cohorts, to identify which factors in childhood (9-13y, Chapter 7) and teenage years (16-20y, Chapter 8) predict adult participation in domestic life and interpersonal relationships. Individuals with limited manual ability, epilepsy or intellectual disability and low levels of mobility capacity, fine motor skills, communication, self-care and social skills are at risk for reduced future participation (See Figure 1, right panel).

The identified predictors on the level of body functions are in line with the available literature on short-term predictors and longitudinal determinants of participation in domestic life and interpersonal relationships.[47-49] In addition, in line with a study on predictors in children with CP, we found that the level of activity and participation at younger age predicted the future outcome of participation in domestic life and interpersonal relationships.[47] Consistent with current practice, this study confirms the importance of addressing gross and fine motor skills in children with cerebral palsy for their future participation, especially in domestic life. In addition, our results suggest that addressing communication, self-care and social skills during pediatric rehabilitation may optimize future participation in interpersonal relationships.

We additionally identified environmental and personal factors predicting adult participation in domestic life and interpersonal relationships. Attending special education, receiving little daily social support and protective or rejective parenting predicted poorer participation in domestic life and interpersonal relationships as an adult. Also, a rigid or avoidant personality and avoidant coping style predicted participation restrictions. Some of these factors of the environment (social support) and personality (behavioral problems) were previously found to be associated to participation in domestic life and interpersonal relationships.[47,50] In a qualitative study, youth with CP highlighted the importance of aspects of the personality and social environment similar to our results in limiting their participation experiences, while a supportive environment, positive coping and personality were supportive factors.[51] Considering environmental and personal factors such as parenting style and personality in rehabilitation with individuals with CP and their families may promote participation. Other contexts than rehabilitation facilities may also address these contextual factors of young individuals with CP, for instance their primary care providers or schools.[52] Future studies may investigate ways to promote a positive social environment and personality or coping of the person. One strategy may be, as was recently suggested,[52] to intervene at promoting inclusion and friendships of children with CP. Additionally, parents may need adequate education on their child's abilities and factors that could support their development. Parent group training was found to reduce behavior problems in children with developmental disabilities and to promote interactions between the parent and child, and may be a suitable approach to improve development of activity and participation for children with CP who experience limitations as well. Finally, future research may aim to identify childhood predictors of adult performance for other domains of activity. Long-term predictors of mobility may be especially interesting, as mobility was established to decline in adults with CP.[53] In addition, families of individuals with CP indicate mobility and self-care as the most important domains of activity and participation.[54] We found that development in these domains is highly related to level of GMFCS. Performance of daily activities in domestic life was found to be predominantly predicted by manual ability and gross motor capacity. Therefore, improving body functions seems to contribute to reaching optimal levels of activities and participation. In addition, personal and environmental factors may be considered in rehabilitation.

METHODOLOGICAL CONSIDERATIONS

Generalizability

Even though a response of 62% is rather high for a 13-year follow-up, the loss-to follow-up of the study may have caused a selection bias. At baseline the cohorts were considered representative of the population of individuals with CP or of the population of individuals with CP of average intelligence (for the oldest cohort). All those known in rehabilitation care belonging to the age cohort in several centers were invited, and we assume that in the Netherlands almost all children with CP are known in rehabilitation care. Loss to follow-up was checked and was not selective regarding sex, GMFCS level and type of CP. Still, we cannot rule out that other factors were related to selection bias. The three youngest cohorts had similar distributions of levels of gross motor function compared to worldwide CP registries.[55] Furthermore, the oldest cohort had a similar distribution of GMFCS levels as a Swedish population-based study after leaving out those with intellectual disability.[27,56]

Some caution is needed when generalizing results to other countries. Participation in some domains, especially domestic life, education, employment, finances and community participation, is influenced by legislation and services, such as those involving social services, sheltered employment and disability benefits. Additionally, the individuals in this study received usual care in the Netherlands, but since it is likely that differences in care and resources affect health issues, activity and participation, results may be different in countries with large differences in healthcare systems or resources. Since we described development of activity and participation in individuals receiving usual care, future research may aim to determine whether differences in timing or intensity of rehabilitation treatment affect this development.

Outcome measures

This study was one of the first to use PROMIS (Patient-Reported Outcomes Measurement Information System) measures in adults with CP. The population had no problems answering these questionnaires, and the measures detected some differences between subgroups of individuals with CP. The reliable and valid PROMIS instruments use T-scores that provide opportunity for comparison to other clinical or geographical populations.[57-62] We used the US population based norms to calculate T-scores to enhance international comparability, which was previously found not to reduce validity in comparison with using Dutch norms in the depression item bank.[57]

In the present studies we used three outcome measures validated to assess self-reported activity and participation performance (VABS), and difficulty (Life-H) and autonomy (RTP) in performance.[45,63,64,66,67] The VABS is a reliable instrument to assess typical development between ages 0-19 years, suitable for individuals with and without disabilities. There may be relevant activities that individuals develop after the ceiling-age and therefore their development may be ongoing although a plateau on the VABS scores is reached. A new version of the questionnaire was adapted to be more suitable for adults,[65] but this version was unavailable at baseline. Future studies may also consider using the Pediatric Evaluation of Disability Inventory Computer Adaptive Test (PEDI-CAT) to assess activity and participation performance in this population. Computer adaptive testing is emerging as an efficient method of assessment, and the PEDI-CAT was recently made suitable for individuals developing into adulthood and validated in individuals with CP.[68]

Finally, intellectual disability showed to be a very important factor determining activity and participation, but was only roughly identified as an IQ below 70, based on education type. Discerning more precisely between different levels of intellectual disability would provide more personalized information for this subgroup of individuals with CP.

CLINICAL RECOMMENDATIONS

Individuals with CP and their families have a need for adequate information on their expected future functioning as a (young) adult with cerebral palsy.[8,69] Questions that are asked to members of the rehabilitation team are for instance '*Will my child be able to live independently in the future?*' or '*Can I have a job and take care of my finances when I grow up?*'. The results from this thesis provide guiding information for subgroups of individuals with CP to questions such as these. To enable adequate knowledge translation, it is necessary to make this information accessible for individuals with CP and their families as well as healthcare providers. Implementation approaches in which products will be developed in co-creation with stakeholders may contribute to adequate implementation of the knowledge.[70,71]

The knowledge provided by this thesis can be used to make some recommendations regarding monitoring of subgroups of individuals with CP in young adulthood. Young adults with CP of average intelligence with GMFCS levels II-V should be monitored regularly, so they can timely receive adequate support when they experience health issues or limitations in activity and participation. In addition, it is important to guide expectations of individuals and their families regarding future functioning in different domains of participation. For individuals of average intelligence and with GMFCS level I the outlook regarding future perceived health, activity and participation is rather positive. In addition to informing them well about possible future restrictions and available treatment programs, it may be suitable for the majority of this subgroup of individuals with CP in their twenties to invite them to contact rehabilitation when they experience health problems. Furthermore, males and those with epilepsy, an avoidant personality or coping style may need support especially, regardless of their level of GMFCS. Interventions may be personalized, according to the domains of activity and participation that are of interest for the individual, and may include transition group programs. Finally, individuals with intellectual disability are a subgroup of individuals with CP in need of specific support.

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SUMMARY

As introduced in **Chapter 1**, cerebral palsy (CP) describes a group of permanent disorders of movement and posture caused by non-progressive disturbances that occurred in the fetal or infant brain. These disorders are often accompanied by other impairments, such as intellectual disability or epilepsy. Nowadays, most individuals with CP are adults, but knowledge gaps exist concerning their functioning and health at adult age. To fill these gaps, we conducted a 13-year follow-up at ages 21-34 years of existing cohorts of children and teens with CP. This thesis described health issues in these young adults with CP and their development of activity and participation into and throughout young adulthood.

In **Chapter 2** we cross-sectionally studied pain, fatigue, depressive symptoms and sleep disturbance in individuals with CP of average intelligence, i.e. those without intellectual disability, in their twenties. Young adults with CP and Gross Motor Function Classification System (GMFCS) level II-V report higher levels of pain and those with GMFCS level III-V report higher levels of fatigue compared to age-matched individuals from the general population, while those with GMFCS level I report lower levels of pain, fatigue and depressive symptoms. Furthermore, pain and fatigue were more strongly inter-related in individuals with CP compared to reference groups, and also related more strongly to global mental health. We therefore recommend to monitor pain and fatigue in young adults with GMFCS levels II-V. Depressive symptoms and sleep disturbance are no prominent health issues in young adults with CP.

In **Chapter 3** we described development curves of performing gross motor, fine motor, self-care, domestic life and community life (finances, employment) activities of individuals with CP from infancy into adulthood per GMFCS level and separately for those with intellectual disability. Development in all domains seemed delayed in individuals with CP compared to reference values of the general population. On average, the maximal gross motor performance level of children with CP was distinctly lower with each lower level of gross motor function. In individuals with CP of average intelligence with walking ability (GMFCS levels I-III) the maximal levels of self-care, domestic life and community life performance did not differ significantly. These individuals approached their limit of gross and fine motor performance at 6-8 years of age, of self-care activities at 11-14 years, of domestic activities at 26-32 years and of community activities at 22-26 years. The ongoing development of daily activities well into adulthood supports the need for taking a life course approach in individuals with CP. Functionally more severely affected individuals (GMFCS IV-V), and those with intellectual disability showed less favourable development of motor performance and daily activities, indicated by much lower maximal levels that they attain at a younger age.

In **Chapter 4** we described development curves of communication and social interactions of individuals with CP from infancy into adulthood per GMFCS level and separately for those with intellectual disability. In communication and social interactions, those of average intelligence showed development curves comparable to references, regardless of their GMFCS level. Thus, we concluded that individuals with CP of average intelligence develop typically in the domains of communication and basic social interactions, despite limitations in motor capacity and activity performance. For these individuals of average intelligence, receptive communication stabilizes in childhood, expressive communication and interpersonal relationships in adolescence and written communication, play & leisure and appropriate social behaviour stabilize in early adulthood. For individuals with intellectual disability, communication and social interactions developed to lower

maximal levels at a younger age, and for individuals with intellectual disability with GMFCS level V maximal levels were even lower. Furthermore, development of communication and social interactions showed large individual variation in individuals with intellectual disability.

In **Chapter 5** the course of difficulty in participation of individuals with CP of average intelligence was studied from adolescence into their early thirties. Although on average they had high levels of participation, a large proportion still experienced difficulties in one or more participation domains, particularly in domestic life, employment and leisure. Difficulty in participation was predominantly experienced by those with lower levels of gross motor function. Moreover, we found in the mid- and late twenties of individuals with CP of average intelligence that difficulty in participation in all domains increased with age, regardless of GMFCS level. This increase in difficulty when individuals grow into adult roles suggests that development towards optimal participation of young adults with CP may be improved.

Autonomy refers to an individual's freedom of decision and ability to act based on own attitudes and reasoning and can be attained in several participation domains. The development of autonomy of individuals with CP from youth into adulthood was described in **Chapter 6**. In most life areas, 90% of individuals with CP reached autonomy in adult roles in their late twenties, with the exception of intimate relationships. Compared to the general population, in intimate relationships individuals with CP were less often autonomous from their teens until their early thirties, and in finances and housing in their (early) twenties. Regardless of age, individuals with GMFCS levels III-V were less often autonomous in transportation and intimate relationships compared to those with GMFCS levels I-II. In the late twenties those with GMFCS levels III-V also became less autonomous in employment and domestic life compared to those with GMFCS levels I-II. The present study was the first to describe the course of autonomy for subgroups of GMFCS levels, and suggested that development of autonomy lagged behind in individuals with CP in intimate relationships, employment and independent living, especially for individuals with lower gross motor function.

Childhood predictors of young adult (21-27 years of age) participation in domestic life and interpersonal relationships were identified in **Chapter 7**. The results showed that childhood factors are highly predictive of participation in young adulthood. Children with CP with limited manual ability, epilepsy, intellectual disability or limited motor capacity were found to be at risk for restricted participation in domestic life and interpersonal relationships in young adulthood. Addressing gross and fine motor skills, communication skills and interpersonal relationships in childhood rehabilitation, especially in individuals at increased risk, might contribute to improved participation in domestic life and interpersonal relationships later in life.

In **Chapter 8** we identified *teenage* predictors of adult (29-34 years of age) participation in domestic life and interpersonal relationships of individuals with CP of average intelligence. Gross motor capacity, environmental factors (parenting style, education type) and personal factors (gender, personality, coping style) were predictors for future participation. Higher levels of gross motor capacity specifically predicted better participation in domestic life. Attending special education, having little daily social support and receiving protective or rejective parenting predicted poorer participation in domestic life and interpersonal relationships. Additionally, being male, a rigid or avoidant personality and avoidant coping style predicted lower future participation in these domains. Finally, the level of communication and self-care skills in teenage life predicted the adult participation outcome. Therefore, adult participation in domestic life and interpersonal relationships may be improved by supporting a positive parenting style and proactive coping ability. Additionally, optimizing gross motor capacity and performance of activities as a teenager may contribute to better participation in domestic life and interpersonal relationships as an adult.

In **Chapter 9** the results of this thesis were discussed in the context of other literature and clinical implications were given. The results of this thesis provided information for subgroups of young individuals with CP based on their GMFCS level and intellectual disability that may guide their expectations for future functioning and health as young adults.

The main messages of this thesis are:

- Young adults with CP of average intelligence with GMFCS levels II-V report much higher levels of pain and slightly higher levels of fatigue than the general population of the same age. Pain and fatigue are highly interrelated and specifically relate to mental health in young adults with CP.
- Individuals with CP of average intelligence continue to develop performance of activities and participation well into adulthood. The development of mobility, self-care, domestic life and community life seems to be delayed for these individuals, while the development of communication and social interaction is comparable to reference values.
- In emerging adults with CP of average intelligence, difficulty in participation in several domains increases and development of autonomy seems to lag behind compared to typically developing youth.
- Individuals with lower levels of gross motor function have lower maximal levels of activity and participation performance, experience more difficulty in participation and are less often autonomous than those with GMFCS level I, especially in domestic life, employment and intimate relationships.
- Individuals with CP with intellectual disability develop to much lower levels of activity and participation performance and reach these levels at younger age.
- Boys, individuals with epilepsy, low mobility capacity, low manual ability, a rigid or avoidant personality, protective or rejective parents and those with low levels of activity and participation in childhood are at increased risk of future participation restrictions.

From the main results of this thesis the following clinical recommendations for subgroups of individuals with CP can be made:

- For young adults with CP of average intelligence with GMFCS level II-V in their twenties regular monitoring of health issues and functioning is suggested, so they can timely receive adequate support.
- For young adults with CP of average intelligence with GMFCS level I in their twenties, on average the outlook regarding future perceived health and performance of activity and participation is rather positive. In addition to informing them well about possible future restrictions and available treatment programs, it may be suitable for the majority of this subgroup of individuals with CP in their twenties to invite them to contact rehabilitation when they experience health problems.
- Males and those with epilepsy, an avoidant personality or coping style may need support especially, regardless of their level of GMFCS.
- For individuals with CP with intellectual disability, support of activity and participation should focus on promoting optimal participation by creating opportunities while offering adequate support and resources.

SAMENVATTING

Zoals geïntroduceerd in **Hoofdstuk 1**, beschrijft cerebrale parese (CP) een groep van permanente beperkingen van houding en beweging die worden veroorzaakt door niet-progressieve verstoringen ontstaan in het brein voor of in het eerste levensjaar. Behalve fysieke beperkingen hebben mensen met CP vaak ook andere stoornissen, zoals bijvoorbeeld een verstandelijke beperking of epilepsie. Alhoewel van alle mensen met CP de grootste groep de volwassen leeftijd heeft bereikt, is er nog altijd weinig kennis over (problemen in) het functioneren als volwassene met CP. Om deze kennishiaten op te vullen is een lange termijn follow-up studie uitgevoerd van cohorten van kinderen en tieners met CP op een leeftijd van 21-34 jaar. Dit proefschrift beschrijft zowel gezondheidsklachten in deze jongvolwassenen als de ontwikkeling en voorspellers van activiteiten en participatie in volwassenheid.

In **Hoofdstuk 2** werden pijn, vermoeidheid, depressieve klachten en slaapproblemen bij twintigers met CP bestudeerd. We vonden dat mensen met Grof Motorisch Functionering Classificatie Systeem (GMFCS) niveau I minder pijn, vermoeidheid, depressieve symptomen en slaapproblemen hadden, terwijl mensen met GMFCS niveaus II-V meer pijn en vermoeidheid hadden dan de algehele bevolking. Bij de mensen met GMFCS niveaus II-V was de ernst van depressieve symptomen en slaapproblemen vergelijkbaar met de algemene bevolking. Daarnaast waren pijn en vermoeidheid sterker aan elkaar gerelateerd bij mensen met CP vergeleken met de algemene bevolking en hadden pijn en vermoeidheid bij hen ook een sterker verband met globale mentale gezondheid. Wij bevelen daarom aan om pijn en vermoeidheid bij jongvolwassenen met GMFCS niveaus II-V te monitoren. Depressieve klachten en slaapproblemen daarentegen behoeven geen extra aandacht bij jongvolwassenen met CP.

In **Hoofdstuk 3** beschreven we de ontwikkeling van de grove motoriek, de fijne motoriek, zelfzorg, het huishoudelijk leven en het maatschappelijk leven (werk, financiële zaken) van mensen met CP van 1 tot 34 jaar. Ontwikkelingscurves werden apart beschreven per GMFCS niveau, en voor mensen met en zonder verstandelijke beperking. De ontwikkeling leek in alle domeinen vertraagd voor mensen met CP in vergelijking met referentiewaarden van de algemene bevolking. Het gemiddelde maximale niveau van het uitoefenen van grof motorische activiteiten was lager bij elk hoger GMFCS niveau, oftewel bij steeds ernstiger beperkt grof motorisch functioneren. Mensen met CP met loopfunctie (GMFCS niveaus I-III) en zonder verstandelijke beperking bereikten gemiddeld 90% van hun maximale niveau van grove en fijne motorische activiteiten tussen 6-8 jaar, van zelfzorg activiteiten tussen 26-32 jaar en van maatschappelijke activiteiten tussen 22-26 jaar. Dat de ontwikkeling van dagelijkse activiteiten doorgaat tot ver in volwassenheid toont de noodzaak van een aanpak voor gezondheidsbevordering over de gehele levensloop van mensen met CP. Ernstig functioneel beperkte mensen (GMFCS niveaus IV-V) en mensen met verstandelijke beperking hadden een minder gunstige ontwikkeling van mobiliteit en dagelijkse activiteiten. Hun ontwikkelingscurves hadden lagere maximale niveaus in alle domeinen, en deze werden bereikt op een jongere leeftijd.

In **Hoofdstuk 4** beschreven we de ontwikkeling van communicatie en sociale interacties van mensen met CP van 1-34 jaar. Ontwikkelingscurves werden apart beschreven per GMFCS niveau en voor mensen met en zonder verstandelijke beperking. Voor de domeinen communicatie en sociale interacties vertoonden mensen met CP zonder verstandelijke beperking vergelijkbare ontwikkeling als de referentiewaarden, onafhankelijk van hun GMFCS niveau. Daarom concludeerden wij dat mensen met CP zonder verstandelijke beperking zich normaal ontwikkelen op de gebieden communicatie en sociale interacties, ondanks beperkingen in motorische capaciteit en dagelijkse activiteiten. De ontwikkeling stabiliseerde voor receptieve communicatie in de kindertijd, voor expressieve communicatie en interpersoonlijke relaties in adolescentie en voor zowel geschreven communicatie, spel en vrije tijd, als sociale vaardigheid in jongvolwassenheid. Mensen met een verstandelijke beperking bereikten op jongere leeftijd een lager maximaal niveau van communicatie en sociale interacties. Hun ontwikkeling vertoonde een grote individuele variatie.

In **Hoofdstuk 5** werd moeite met participatie onderzocht bij mensen met CP zonder verstandelijke beperking tussen 16 en 34 jaar oud. Hoewel zij gemiddeld op een hoog niveau participeerden, ervoer een groot deel moeite in één of meer participatiedomeinen, vooral in huishoudelijk leven, werk en recreatie. Moeite met participatie kwam vooral voor bij mensen met lagere niveaus van grof motorisch functioneren. Daarnaast nam bij twintigers met CP zonder verstandelijke beperking de moeite met participatie toe in alle domeinen, onafhankelijk van het GMFCS niveau. Deze resultaten suggereren dat de ontwikkeling van optimale participatie van jongvolwassenen met CP kan worden verbeterd, vooral voor mensen met lagere niveaus van grof motorisch functioneren.

Autonomie verwijst naar de keuzevrijheid en onafhankelijkheid in handelen van een individu, gebaseerd op iemands eigen attitudes en redeneringen. In **Hoofdstuk 6** werd de ontwikkeling van autonomie in verschillende domeinen van participatie beschreven voor mensen met CP van 12-34 jaar. Deze studie beschreef voor het eerst de ontwikkeling van autonomie voor subgroepen van mensen met CP gebaseerd op GMFCS niveaus. In de meeste levensgebieden ontwikkelde 90% van de twintigers met CP naar autonomie in volwassen rollen, behalve in intieme relaties: vergeleken met de algehele bevolking hadden mensen met CP minder vaak intieme relaties. In financieel en huishoudelijk leven waren twintigers met CP ook minder vaak autonoom. Ongeacht leeftijd waren mensen met GMFCS niveaus III-V minder vaak autonoom in transport en intieme relaties in vergelijking met mensen met GMFCS niveaus I-II. Boven de 25 waren mensen met GMFCS niveau III-V ook minder vaak autonoom in werk en huishoudelijk leven dan mensen met GMFCS niveaus I-II. In conclusie, in vergelijking met de algemene bevolking loopt de ontwikkeling van autonomie bij mensen met CP achter op het gebied van intieme relaties, werk en huishoudelijk leven. Mensen met GMFCS niveaus III-V worden minder vaak autonoom in deze gebieden.

We identificeerden factoren uit de kindertijd die participatie in huishoudelijk leven en interpersoonlijke relaties in jongvolwassenheid voorspellen in **Hoofdstuk 7**. Kinderen met CP met beperkte manuele vaardigheden, epilepsie, verstandelijke beperking of verminderde motorische capaciteit bleken als jongvolwassenen vaker beperkingen in het huishoudelijk leven en interpersoonlijke relaties te hebben. Daarnaast voorspelden het niveau van fijn motorische activiteiten, communicatie en interpersoonlijke relaties als kind de mate van participatie als jongvolwassene. Het verbeteren van fijne motoriek, communicatie en interpersoonlijke relaties in de kinderrevalidatie zou daarom kunnen bijdragen aan verbeterde participatie op latere leeftijd.

In **Hoofdstuk 8** identificeerden we voorspellers uit de tienertijd voor participatie in huishoudelijk leven en interpersoonlijke relaties voor volwassen mensen met CP zonder verstandelijke beperking. Grof motorische capaciteit, omgevingsfactoren (opvoedstijl, schooltype) en persoonlijke factoren (geslacht, persoonlijkheid, coping stijl) als tiener bleken belangrijke voorspellende factoren voor participatie als volwassene. Hoe beter de grof motorische capaciteit hoe beter de participatie in het huishoudelijk leven. Als iemand speciaal onderwijs had gevolgd, weinig dagelijkse sociale steun had ontvangen of een beschermende of afwijzende opvoeding had genoten, voorspelde dat verminderde participatie in huishoudelijk leven en interpersoonlijke relaties. Daarnaast participeerden zowel mannen als mensen met een rigide of vermijdende persoonlijkheid en een vermijdende coping stijl minder in deze domeinen. Tenslotte waren communicatie en zelfzorgactiviteiten als tiener gerelateerd aan de participatie in huishoudelijk leven en interpersoonlijke relaties als volwassene. Het lijkt er daarom op dat participatie in huishoudelijk leven en interpersoonlijke relaties verbeterd zou kunnen worden door het bevorderen van een positieve opvoedstijl en proactieve coping stijl. Verder kan het optimaliseren van grof motorische capaciteit en dagelijkse activiteiten in de tienerleeftijd ervoor zorgen dat iemand als volwassene beter participeert in het huishoudelijke leven en interpersoonlijke relaties.

In **Hoofdstuk 9** bediscussieerden we de resultaten van dit proefschrift in de context van andere literatuur en gaven we klinische implicaties. Hieruit kwam algemene informatie over het functioneren en mogelijke problemen van subgroepen jongvolwassenen met CP voort, die richting kan geven aan de verwachtingen van jonge mensen met CP.

De belangrijkste boodschappen van dit proefschrift zijn:
- Jongvolwassenen met CP zonder verstandelijke beperking met GMFCS niveaus II-V hebben ernstigere pijn en licht verhoogde niveaus van vermoeidheid in vergelijking met de algehele bevolking. Pijn en vermoeidheid zijn sterk aan elkaar gerelateerd en relateren in het bijzonder aan de mentale gezondheid van jongvolwassenen met CP.
- Mensen met CP zonder verstandelijke beperking ontwikkelen zich in het uitvoeren van activiteiten en participatie tot ver in hun volwassenheid. Deze ontwikkeling is vertraagd voor motorisch functioneren, zelfzorg, huishoudelijk en maatschappelijk leven, maar vergelijkbaar met referentiewaarden voor communicatie en sociale interacties.
- Tijdens de ontwikkeling naar volwassenheid neemt moeite met participatie in verschillende domeinen toe bij mensen met CP zonder verstandelijke beperking, en lijken zij achter te lopen in de ontwikkeling naar autonomie.
- Mensen met CP met lagere niveaus van grof motorisch functioneren hebben lagere maximale niveaus van participatie, ervaren meer moeite in participatie en zijn minder vaak autonoom dan mensen met een hoog niveau van grof motorisch functioneren, met name in de domeinen huishoudelijk leven, werk en intieme relaties.
- Mensen met CP met verstandelijke beperking ontwikkelen tot lagere maximale niveaus van activiteiten en participatie, en bereiken deze reeds op jonge leeftijd.
- Jongens, mensen met epilepsie, lagere manuele capaciteiten, een rigide of vermijdende persoonlijkheid en beschermende of afwijzende ouders hebben een verhoogd risico op beperkingen in participatie in volwassenheid.

Deze resultaten van dit proefschrift leiden tot de volgende klinische aanbevelingen voor subgroepen van mensen met CP:
- Voor twintigers met CP zonder verstandelijke beperking met GMFCS niveaus II-V stellen we regelmatige monitoring voor, zodat tijdig passende ondersteuning kan worden ingezet.
- Voor twintigers met CP zonder verstandelijke beperking met GMFCS niveau I is de ervaren gezondheid en uitvoeren van activiteiten en participatie gemiddeld redelijk positief. Behalve het zorgvuldig informeren over mogelijke problemen in de toekomst en beschikbare behandelmethoden, lijkt het passend voor de meerderheid van deze groep om zelf contact op te nemen met de revalidatie wanneer zij problemen ervaren.

- Mannen, mensen met epilepsie, lagere manuele vaardigheden, een vermijdende persoonlijkheid en beschermende of afwijzende ouders hebben mogelijk meer ondersteuning nodig, onafhankelijk van hun niveau van GMFCS.
- Om activiteiten en participatie van mensen met CP met een verstandelijke beperking te ondersteunen is het raadzaam om te concentreren op het creëren van mogelijkheden tot deelnemen, en het tegelijkertijd bieden van voldoende steun en middelen.

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- Leden van de leescommissie
- Collega's van het VUmc en Erasmus MC
- Familie, vrienden en Vishal

Heel erg bedankt!

CURRICULUM VITAE

Marloes van Gorp was born on the 7th of May 1989 in Best, the Netherlands. She attended the Gymnasium at Pleincollege Bisschop Bekkers in Eindhoven and gruaduated in 2007 with a specialization in nature and health. She advanced to study health sciences at Maastricht University. During the course of the first year, she became aware that a practical education might fit her better and she switched to study physiotherapy. In parallel, she attended the premaster course of physiotherapy science at the University of Utrecht, where she first got in contact with research. In 2011 she received her Bachelor degree in physiotherapy from the University of Applied Science Utrecht. She started working as a physiotherapist in general practices, but primarily in elderly rehabilitation.

To gain further understanding of human movement and research, and to be able to contribute to improvement of the field of rehabilitation, she followed the health track of the master Human Movement Sciences: Sport, Exercise and Health at the VU University Amsterdam. She did her research internship at the Anton de Kom University in Paramaribo, Suriname. Her thesis, called 'Health-related fitness, motor coordination, physical and sedentary activities of urban and rural children in Suriname', received a 9/10 and was later published in the peer-reviewed *Journal of Physical Activity and Health*. She obtained her Master degree cum laude in 2013.

In 2015 she took on the position of junior researcher at the departments of rehabilitation medicine at the VU University Medical Center and Erasmus University Medical Center. She was appointed to a long-term follow-up study of the Pediatric Rehabilitation Research in the Netherlands consortium, focusing on individuals with CP. After a swift period of data-collection, the combination of follow-up and available data provided plenty opportunity to analyze and write about health, activity and participation of young adults with CP. The results of her committed work were described in this thesis.

LIST OF PUBLICATIONS

This thesis is based on the following international peer-reviewed publications:
Van Gorp M, Roebroeck ME, van Eck M, Voorman JM, Twisk JWR, Dallmeijer AJ, van Wely L. Childhood factors predict participation of young adults with cerebral palsy in domestic life and interpersonal relationships: a prospective cohort study. *Disability and Rehabilitation.* 2019.

Van Gorp M, van Wely L, Dallmeijer AJ, de Groot V, Ketelaar M, Roebroeck ME, PERRIN-DECADE study group. Long-term course of difficulty in participation of individuals with cerebral palsy aged 16 to 34 years: a prospective cohort study. *Developmental Medicine & Child Neurology.* 2019; 61(2)194-203.

Van Gorp M, Roebroeck ME, Tan SS, de Groot V, Gorter JW, Smits DW, Schmidt AK, Dallmeijer AJ, PERRIN DECADE study group. Activity performance curves of individuals with cerebral palsy. *Pediatrics*. 2018; 142(5)e20173723.

Tan SS, van Gorp M, Voorman JM, Geytenbeek JJM, Reinders-Messelink HA, Ketelaar M, Dallmeijer AJ, Roebroeck ME, on behalf of the PERRIN DECADE study group. Development curves of communication and social interactions in cerebral palsy. *Developmental Medicine & Child Neurology*. In production. **Joint first author**.

Schmidt AK, van Gorp M, van Wely L, Ketelaar M, Hilberink SR, Roebroeck ME, on behalf of the PERRIN DECADE study group. Autonomy in participation of individuals with cerebral palsy from youth into adulthood. Submitted. **Joint first author**.

Van Wely L, van Gorp M, Tan SS, van Meeteren J, Roebroeck ME, Dallmeijer AJ on behalf of the PERRIN DECADE study group. Teenage predictors of participation of adults with cerebral palsy in domestic life and interpersonal relationships: a 13-year follow-up study. Submitted.

Van Gorp M, Dallmeijer AJ, van Wely L, de Groot V, Terwee CB, Flens G, Stam HJ, van der Slot W, Roebroeck ME, on behalf of the PERRIN DECADE study group. Pain, fatigue, depressive symptoms and sleep disturbance in young adults with cerebral palsy. Submitted.

Other international peer-reviewed publications by the author:
Walhain F, van Gorp M, Lamur KS, Veeger HEJ, Ledebt A. Health-related fitness, motor coordination, and physical and sedentary activities of urban and rural children in Suriname. *Journal of Physical Acitivty and Health*. 2016; 13(10)1035-1041.

Smits DW, van Gorp M, van Wely L, Verheijden J, Voorman JM, Wintels S, van der Cruijsen J, Ketelaar M. Participation in social roles of adolescents with cerebral palsy: exploring accomplishment and satisfaction. *Archives of Rehabilitation Research & Clinical Translation.* In production.

Contributions to international scientific conferences:
Van Gorp M, Dallmeijer AJ, de Groot V, Terwee CB, Flens G, Stam HJ, van der Slot W, Roebroeck ME, on behalf of the PERRIN DECADE study group. Comparing pain, fatigue, depressive symptoms and sleep disturbance between young adults with cerebral palsy and a reference population. Oral presentation at the 73rd AACPDM 2019, Anaheim.

Van Gorp M, Tan SS, Voorman JM, Geytenbeek JJM, Reinders-Messelink HA, Ketelaar M, Dallmeijer AJ, Roebroeck ME. Development curves of communication and social interactions of individuals with cerebral palsy. Oral presentation at the 73rd AACPDM 2019, Anaheim.

Van Gorp M, van Wely L, Dallmeijer AJ, van der Slot W, de Groot V, Stam HJ, Roebroeck ME. Pain, fatigue and sleep disturbances in young adults with cerebral palsy. Oral presentation at the 31st EACD 2019, Paris. *Developmental Medicine & Child Neurology*. 2019; 61(S2) 41.

Van Gorp M, Roebroeck ME, van Eck M, Voorman J, Twisk JWR, Dallmeijer AJ, van Wely L. Childhood factors predict participation of young adults with cerebral palsy in domestic life and interpersonal relationships: a prospective cohort study. Poster presentation at the 31st EACD 2019, Paris.

Van Gorp M, Roebroeck ME, van Wely L, de Groot V, Gorter JW, Smits DW, Schmidt AK, Dallmeijer AJ. Development curves of motor and daily activity performance of individuals with cerebral palsy from childhood into adulthood. Poster presentation at the 72nd AACPDM 2018, Cincinnati. *Developmental Medicine & Child Neurology*. 2018; 60(S3) 87-88.

Van Gorp M, Roebroeck ME, van Wely L, de Groot V, Gorter JW, Smits DW, Schmidt AK, Dallmeijer AJ. Development curves of motor and daily activity performance of individuals with cerebral palsy from childhood into adulthood. Oral presentation at the 12th ISPRM 2018, Paris. *Annals of Physical and Rehabilitation Medicine*. 2018; 61(supplement) e54-e55.

Van Gorp M, van Wely L, Dallmeijer AJ, de Groot V, Ketelaar M, Roebroeck ME. Difficulty and independence in participation of emerging adults with cerebral palsy. Poster presentation at the 12th ISPRM 2018, Paris. *Annals of Physical and Rehabilitation Medicine*. 2018; 61(supplement) e307.

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Van Gorp M, Roebroeck ME, van Wely L, de Groot V, Gorter JW, Smits DW, Schmidt AK, Dallmeijer AJ. Development of performance of gross and fine motor skills in individuals with cerebral palsy. Joint congress of the DCRM, BNF-PRM and RBSPRM 2017, Maastricht. Oral presentation at the Joint congress of the DCRM, BNF-PRM and RBSPRM 2017. *Journal of Rehabilitation Medicine*. 2018; 50 102-103.

Van Gorp M, van Wely L, Dallmeijer AJ, de Groot V, Ketelaar M, Roebroeck ME. Difficulty and independence in participation during transition into adulthood: a prospective cohort study of individuals with cerebral palsy. Oral presentation at the Joint congress of the DCRM, BNF-PRM and RBSPRM 2017, Maastricht. *Journal of Rehabilitation Medicine.* 2018; 50 103.

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Van Wely L, van Gorp M, van Meeteren J, Roebroeck ME, Dallmeijer AJ. Early predictors of participation of adults with cerebral palsy in interpersonal relationships. Oral presentation at the 29th EACD 2017, Amsterdam. *Developmental Medicine & Child Neurology*. 2017; 59(S2) 33.

Tan SS, van Gorp M, van Wely L, Smits, DW, van der Slot WM, Ketelaar M, Dallmeijer, AJ, Roebroeck ME. A decade later: developmental trajectories of social participation and associated factors in individuals with cerebral palsy into adulthood. Poster presentation at the EACD 2017, Amsterdam. *Developmental Medicine & Child Neurology*. 2017; 59(S2) 60.

Awards:

Nominated for Gayle G Arnold award, AACPDM 2019
AACPDM Student Travel Scholarship, AACPDM 2018, AACPDM 2019
Erasmus Trustfonds Travel Grant, ISPRM 2018

PHD PORTFOLIO

Summary of PhD training and teaching activities

Name PhD student: Marloes van Gorp
Erasmus MC Department: Rehabilitation Medicine
Amsterdam UMC, location VUmc department: Rehabilitation Medicine
Research School: AMS/NIHES

PhD period: 2015-2019
Promotor(s): Prof. H.J. Stam, Prof. V. de Groot
Supervisor: Dr. M.E. Roebroeck, Dr. A.J. Dallmeijer

1. PhD training

	Year	Workload (ECTS)
General academic skills		
- Basic course for clinical investigators (BROK), NFU, Amsterdam	2015	1.5
- Writing a scientific article, Taalcentrum VU, Amsterdam	2017	3
- Scientific integrity, Erasmus MC, Rotterdam	2019	0.3
- Masterclass grant writing for ZonMw, Erasmus MC, Rotterdam	2019	0.3
- Reference manager, Medical library, VUmc, Amsterdam	2015	0.2
Research skills		
- Regression techniques, EpidM, Amsterdam	2015	5
- Longitudinal data-analysis, EpidM, Amsterdam	2016	3
Presentations		
- Oral presentation at the 29th EACD.	2017	0.5
- Two oral presentations at the Joint congress of the DCRM, BNF-PRM and RBSPRM.	2017	1.0
- Poster presentation at the 12th ISPRM.	2018	0.3
- Oral presentation at the 12th ISPRM.	2018	0.5
- Poster presentation at the 72nd AACPDM.	2018	0.3
- Oral presentation at the 31st EACD.	2019	0.5
- Poster presentation at the 31st EACD.	2019	0.3
- Two oral presentations at the 73rd AACPDM.	2019	1.0
- Oral presentation: 'PERRIN DECADE onderzoeksopzet' Regional meeting for rehabilitation physicians, Rotterdam	2015	0.3
- Oral presentation: 'Childhood predictors of domestic life and interpersonal relationships of young adults with cerebral palsy' Regional meeting for rehabilitation physicians, Rotterdam.	2018	0.3
- Oral presentation: 'Performance curves of individuals with cerebral palsy' Regional meeting for rehabilitation physicians, Rotterdam.	2019	0.3
- Oral presentation: 'Performance curves of individuals with cerebral palsy' Regional meeting for rehabilitation physicians, Amsterdam.	2019	0.3
- Oral presentations: Several topics. Research meetings dept. of Rehabilitation Medicine Erasmus MC and Amsterdam UMC, location VUmc, Rotterdam and Amsterdam.	2015-2019	1

	Year	**Workload (ECTS)**
International conferences and symposia		
- 29th European Academy of Childhood Disability Conference, Amsterdam, The Netherlands	2017	1.2
- Joint Congress of the DCRM, BNF-PRM and RBSPRM, Maastricht, The Netherlands	2017	0.6
- 12th International Society of Physical and Rehabilitation Medicine World Congress, Paris, France	2018	1.2
- 72nd Annual Meeting of the American Academy for Cerebral Palsy and Developmental Medicine, Cincinnati, Ohio, USA	2018	1.2
- 31s European Academy of Childhood Disability Conference, Paris, France	2019	1.2
- Combined 73rd Annual Meeting of the American Academy for Cerebral Palsy and Developmental Medicine and 2nd Triannual International Alliance of Academies of Childhood Disability meeting, Anaheim, California, USA	2019	1.2
Seminars and workshops		
- Attending symposium: Cerebrale parese op volwassen leeftijd, Rijndam Revalidatie/Erasmus MC, Rotterdam	2018	0.2
- Presenter in instructional course: Long-term development and early predictors of participation of adults with CP in domestic life and interpersonal relationships: Implementation using co-creation. 72nd AACPDM.	2018	0.5
- Presenter in mini-symposium: Long-term development into adulthood and personal experiences of participation of individuals with CP: the Dutch PERRIN study. 31st EACD.	2019	0.5
- Presenter at meeting Pediatric rehabilitation physicians of Netherlands Society of Rehabilitation Medicine: PERRIN 2.0., Utrecht.	2019	0.3
- Presenter in breakfast session: Characterizing developmental stages in individuals with cerebral palsy: from mobility to social interactions. 73rd AACPMD.	2019	0.5
Didactic skills		
- ‘Docentenopleiding HBO-docent medisch-biologische vakken’, Vrije Universiteit, Amsterdam	2017-2018	22.5
Other		
- Organizing VVBN PhD-day, Erasmus MC, Rotterdam	2017	0.5
- Attending VVBN PhD-day, Vrije Universiteit, Amsterdam	2018	0.2
- Attending AMS annual meeting, Vrije Universiteit, Amsterdam	2019	0.2
- Attending research meetings of the departments	2015-2019	6

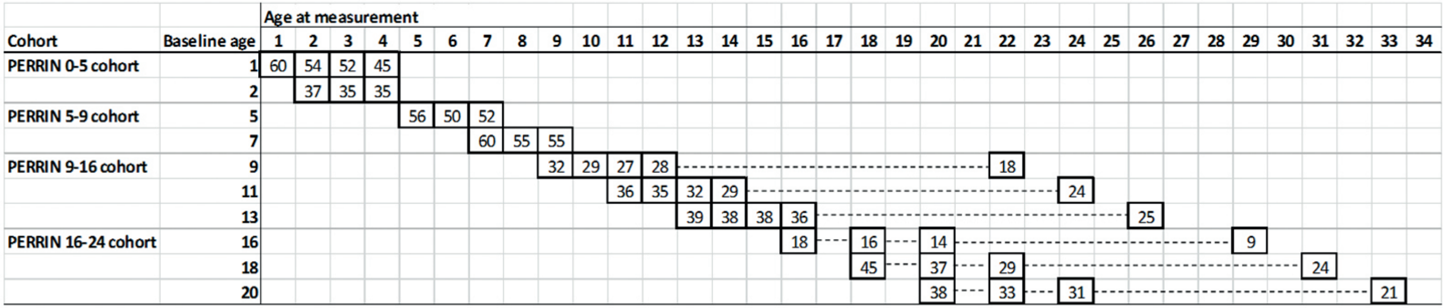
2. Teaching activities

	Year	**Workload (Hours/ ECTS)**
Lecturing		
- Lecturing werkgroepen keuze onderwijs wetenschap, bachelor of medicine, Vrije Universiteit, Amsterdam	2016	1
- Teaching internship, Utrecht University of Applied Science, Utrecht	2017-2018	7.5
- Second assessor theses of Bachelor of Medicine, Vrije Universiteit, Amsterdam	2019	1
Supervision of students		
- Supervising a bachelor thesis in Medicine	2017	1
- Supervising of a MD in residency	2018	0.5
Total	2015-2019	68.9

ONLINE SUPPLEMENTAL INFORMATION

Chapter 3	Activity performance curves of individuals with CP
Chapter 4	Development curves of communication and social interaction in cerebral palsy
Chapter 5	Long-term course of difficulty in participation of individuals with cerebral palsy aged 16-34 years: a prospective cohort study
Chapter 6	Autonomy in participation of individuals with cerebral palsy from youth into adulthood

SUPPLEMENTAL INFORMATION CHAPTER 3



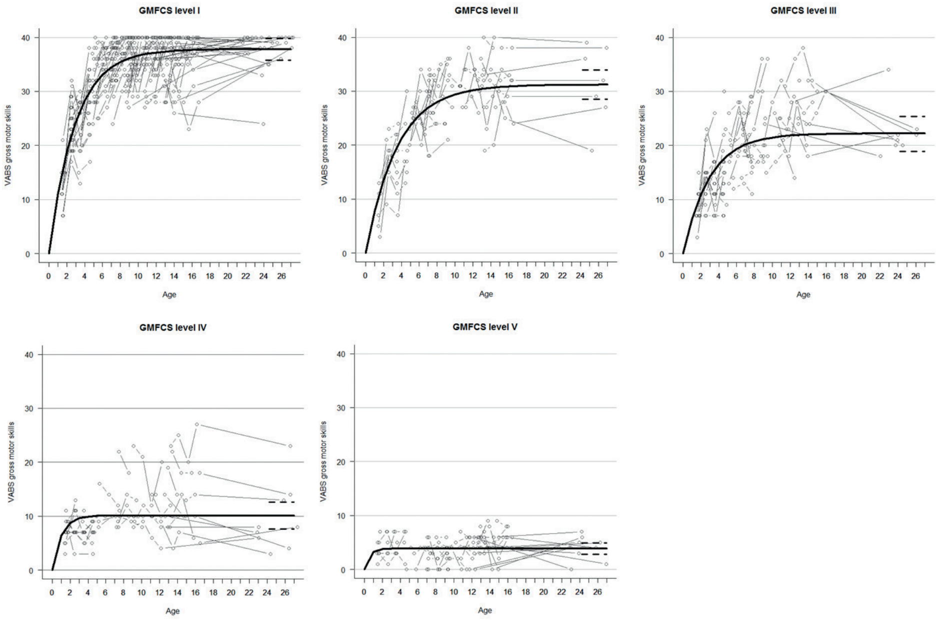
		Age at measurement																																	
Cohort	**Baseline age**	**1**	**2**	**3**	**4**	**5**	**6**	**7**	**8**	**9**	**10**	**11**	**12**	**13**	**14**	**15**	**16**	**17**	**18**	**19**	**20**	**21**	**22**	**23**	**24**	**25**	**26**	**27**	**28**	**29**	**30**	**31**	**32**	**33**	**34**
PERRIN 0-5 cohort	**1**	60	54	52	45																														
	2		37	35	35																														
PERRIN 5-9 cohort	**5**					56	50	52																											
	7							60	55	55																									
PERRIN 9-16 cohort	**9**									32	29	27	28										18												
	11											36	35	32	29										24										
	13													39	38	38	36										25								
PERRIN 16-24 cohort	**16**																18		16		14									9					
	18																		45		37		29									24			
	20																				38		33		31									21	

Appendix 1: Number of observations by age group (in years).

Appendix 2: Average development curves (dotted lines: 50% ranges of the limit) with the raw observations indicated in gray.

A. Gross motor performance development curves for GMFCS levels I-V
B. Fine motor performance development curves for MACS levels I-V
C. Personal daily activity development curves for GMFCS levels I through IV individuals no ID and GMFCS levels I-III and IV-V for individuals with ID
D. Domestic daily activity development curves for GMFCS levels I through IV individuals no ID and GMFCS levels I-III and IV-V for individuals with ID
E. Community daily activity development curves for GMFCS levels I through IV individuals no ID and GMFCS levels I-III and IV-V for individuals with ID

A.



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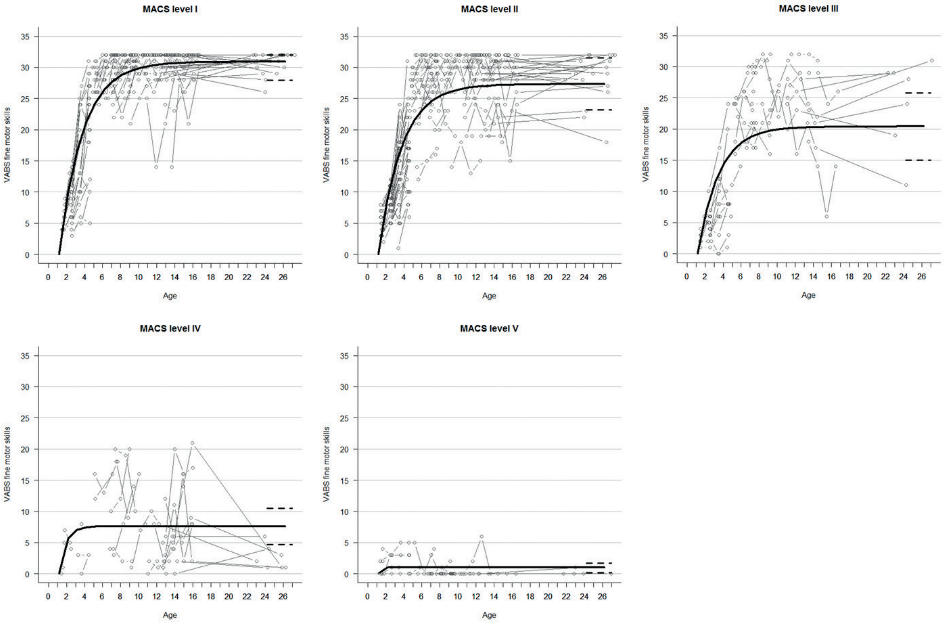
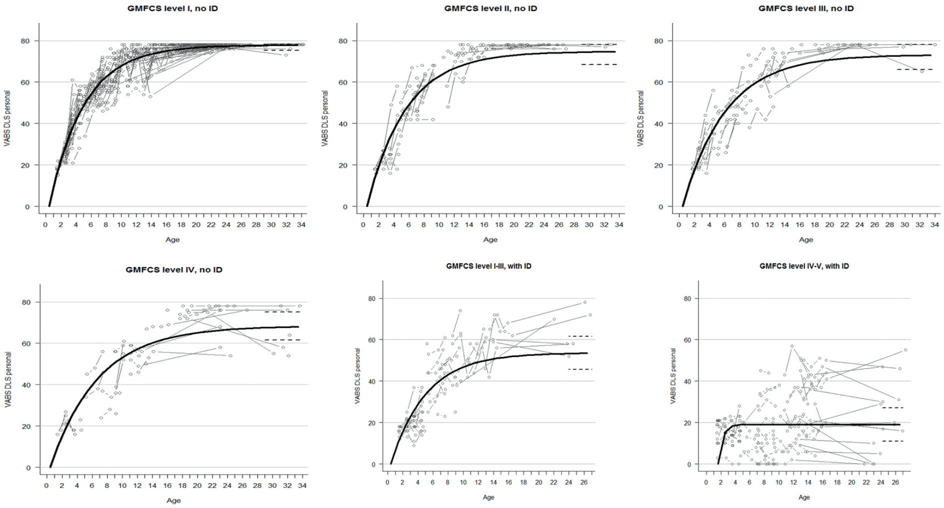

MACS level I
MACS level II
MACS level III
MACS level IV
MACS level V
VABS fine motor skills
0
5
10
15
20
25
30
35
Age
0 2 4 6 8 10 12 14 16 18 20 22 24 26

C.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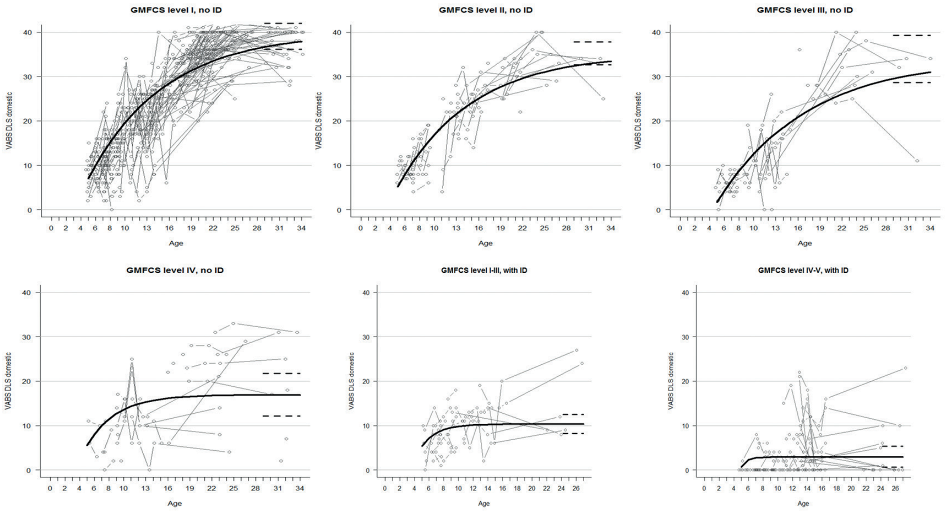
GMFCS level III, no ID
GMFCS level IV, no ID
GMFCS level I-III, with ID
GMFCS level IV-V, with ID
VABS DLS personal
0
20
40
60
80
Age
0 2 4 6 8 10 12 14 16 18 20 22 24 26 28 30 32 34

D.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GMFCS level II, no ID
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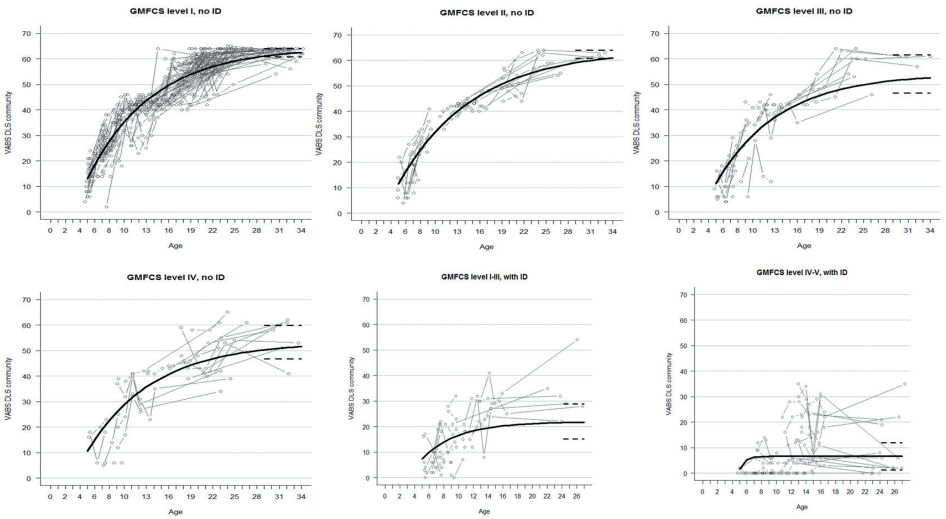
GMFCS level IV, no ID
GMFCS level I-III, with ID
GMFCS level IV-V, with ID
VABS DLS domestic
0
10
20
30
40
Age
0 2 4 6 8 10 13 16 19 22 25 28 31 34
0 2 4 6 8 10 12 14 16 18 20 22 24 26

E.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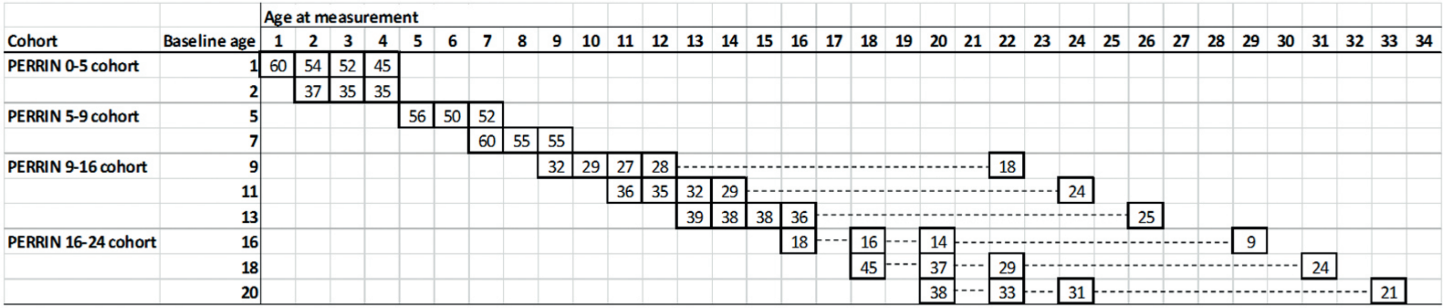
GMFCS level II, no ID
GMFCS level III, no ID
GMFCS level IV, no ID
GMFCS level I-III, with ID
GMFCS level IV-V, with ID
VABS DLS community
0
10
20
30
40
50
60
70
Age
0 2 4 6 8 10 13 16 19 22 25 28 31 34
0 2 4 6 8 10 12 14 16 18 20 22 24 26

SUPPLEMENTAL INFORMATION CHAPTER 4



		Age at measurement																																	
Cohort	**Baseline age**	**1**	**2**	**3**	**4**	**5**	**6**	**7**	**8**	**9**	**10**	**11**	**12**	**13**	**14**	**15**	**16**	**17**	**18**	**19**	**20**	**21**	**22**	**23**	**24**	**25**	**26**	**27**	**28**	**29**	**30**	**31**	**32**	**33**	**34**
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	11											36	35	32	29										24										
	13													39	38	38	36										25								
PERRIN 16-24 cohort	**16**																18		16		14									9					
	18																		45		37		29									24			
	20																				38		33		31									21	

Appendix 1: Number of observations by age group

Appendix 2: Statistical appendix

To estimate limit and rate parameters for each subdomain of communication and social interactions, a nonlinear mixed-effects model was used on the subdomain scores, separately for subgroups of individuals in different GMFCS levels and individuals without or with ID. The model used is similar to the stable limit model used by Rosenbaum and colleagues and Smits and colleagues (2002)[12] and explained in the statistical appendix of Hanna and colleagues.[3] We adjusted this model, adding an offset (start age of development) from the origin for those domains where development started at a later age (i.e. not in infancy).

Within the nlme package of R we defined an asymptotic regression model (inverse exponential function) from the start age of development. This start age (x_0) was separately estimated from the data. Thus, communication and social interaction subdomains (y) were modeled as a function of age in years (x):

$y = L(1\text{-}\exp[-R(x\text{-}x_0)])$

In this function, L describes the asymptote parameter, which is the value toward which the curve levels off at high age, referred to as the limit of development of communication and social interaction. The R parameter describes the rate of development towards this limit. To keep this parameter positive during optimization, R was substituted for exp[logR], so the estimated parameter, logR, is the natural log of the rate. Finally, x_0 was the predetermined offset, or start age of development, that provided the best model fit for each subdomain according to the Akaike Information Criterion when using all observations of all individuals (GMFCS level I-V, without and with ID) of the relevant subdomain. With respect to the coping subdomain, the model for individuals with ID and GMFCS-level V did not fit using x_0 determined based on all observations. Therefore, x_0 for this subgroup was based on the observations of individuals with ID and GMFCS level V only.

To aid interpretability of the rate parameter, it was transformed to age_{90}, i.e. the average age at which individuals reached 90% (0.9 in the below formula) of their limit. The following transformation (adjusted from Hanna and colleagues) was used:

$\text{Age}_{90} = x_0 + \ln(1/[1-0.9])/\exp(\log R)$

---

1. Hanna SE, Rosenbaum PL, Bartlett DJ, et al. Stability and decline in gross motor function among children and youth with cerebral palsy aged 2 to 21 years. *Dev Med Child Neurol.* 2009;51(4):295-302.
2. Smits DW, Gorter JW, Riddell CA, et al. Mobility and self-care trajectories for individuals with cerebral palsy (aged 1-21 years): a joint longitudinal analysis of cohort data from the Netherlands and Canada. Lancet Child Adolesc Health 2019 doi: 10.1016/S2352-4642(19)30122-1
3. Rosenbaum PL, Walter SD, Hanna SE, et al. Prognosis for gross motor function in cerebral palsy: creation of motor development curves. JAMA. 2002;288(11):1357-1363.

Appendix 3: Average development curves (dotted lines: 50% ranges of the limit) with the raw observations indicated in gray.

A. Receptive communication development curves for GMFCS levels I through IV individuals no ID and GMFCS levels I-III and IV-V for individuals with ID

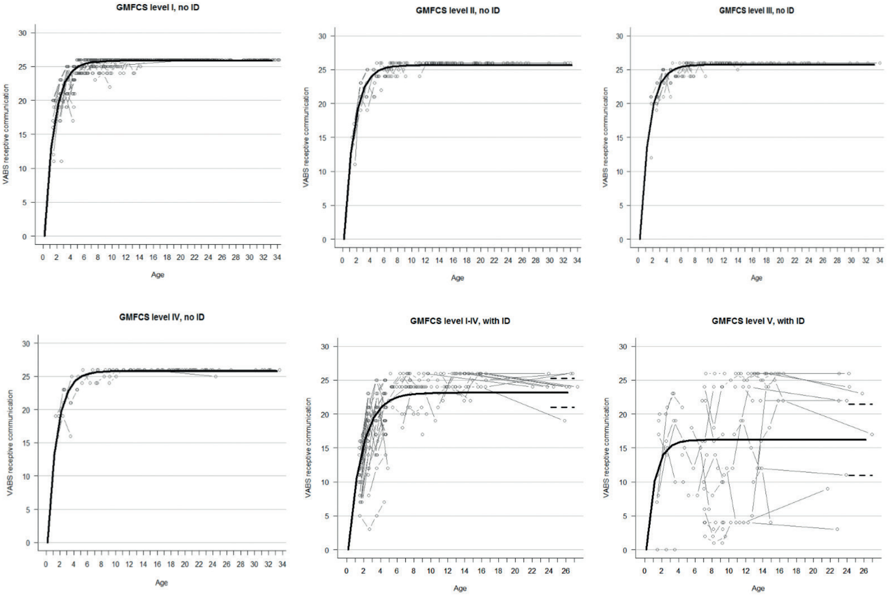
B. Expressive communication development curves for GMFCS levels I through IV individuals no ID and GMFCS levels I-III and IV-V for individuals with ID C. Written communication development curves for GMFCS levels I through IV individuals no ID and GMFCS levels I-III and IV-V for individuals with ID

D. Interpersonal relationships development curves for GMFCS levels I through IV individuals no ID and GMFCS levels I-III and IV-V for individuals with ID

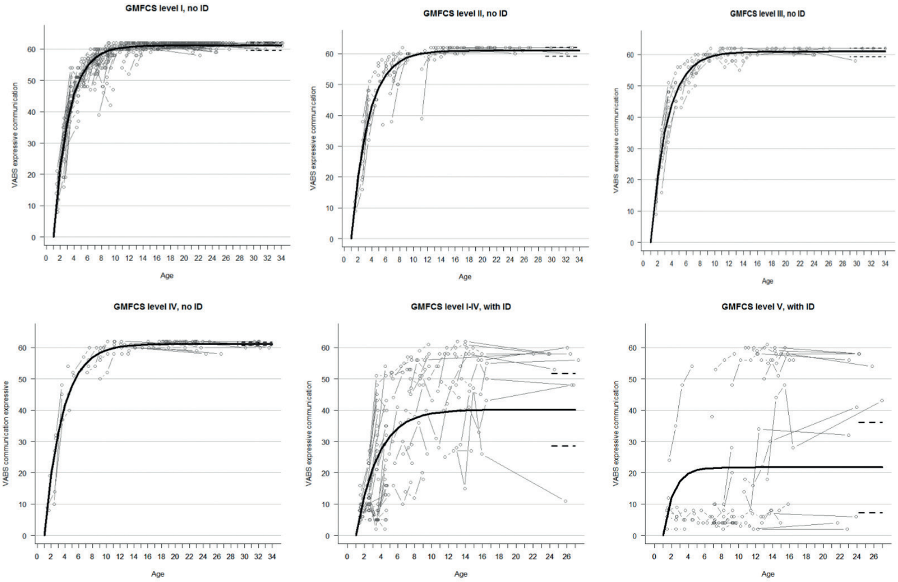
E. Play and leisure development curves for GMFCS levels I through IV individuals no ID and GMFCS levels I-III and IV-V for individuals with ID

F. Coping development curves for GMFCS levels I through IV individuals no ID and GMFCS levels I-III and IV-V for individuals with ID

A.



B.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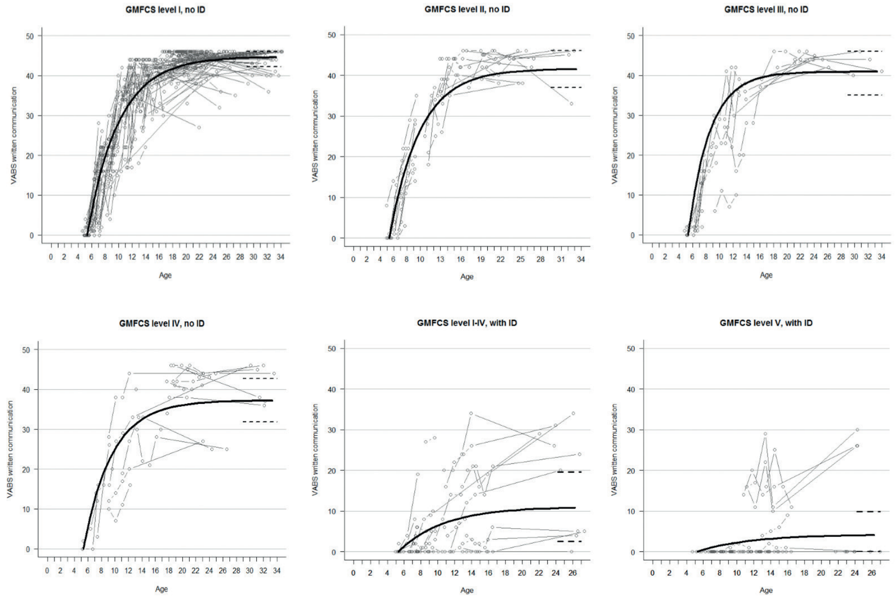
GMFCS level III, no ID
GMFCS level IV, no ID
GMFCS level I-IV, with ID
GMFCS level V, with ID
VABS expressive communication
VABS communication expressive
Age
0
10
20
30
40
50
60
0 2 4 6 8 10 12 14 16 18 20 22 24 26 28 30 32 34
0 2 4 6 8 10 12 14 16 18 20 22 24 26

C.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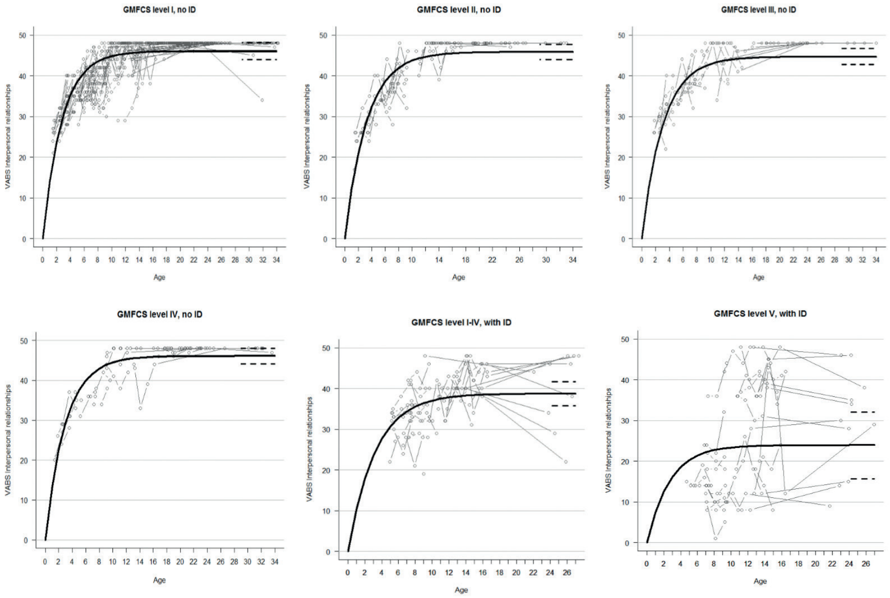
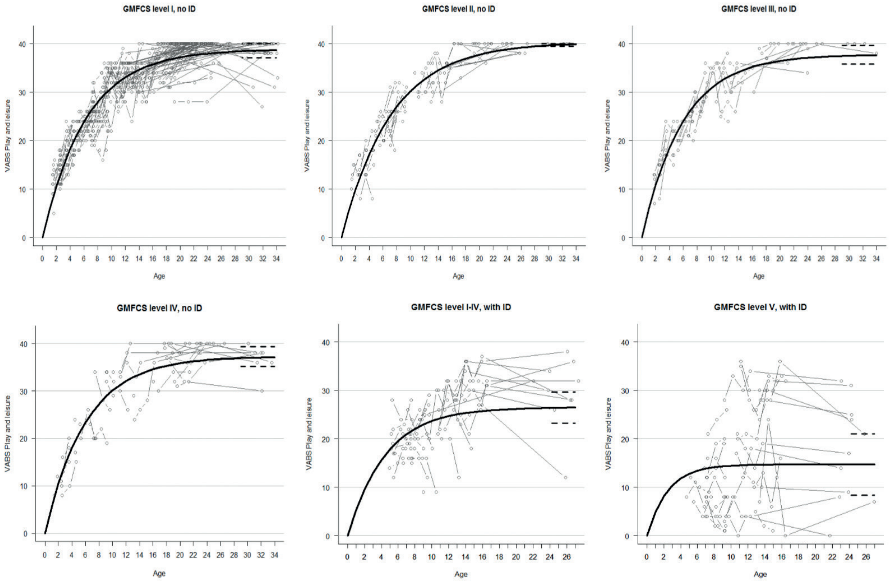
GMFCS level II, no ID
GMFCS level III, no ID
GMFCS level IV, no ID
GMFCS level I-IV, with ID
GMFCS level V, with ID
VABS written communication
Age
0
10
20
30
40
50
0 2 4 6 8 10 12 14 16 18 20 22 24 26 28 30 32 34
0 2 4 6 8 10 13 16 19 22 25 28 31 34
0 2 4 6 8 10 12 14 16 18 20 22 24 26

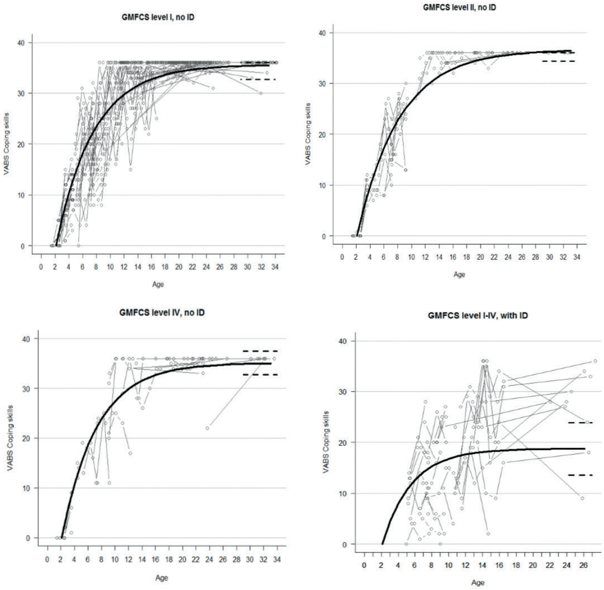
D.


GMFCS level I, no ID
GMFCS level II, no ID
GMFCS level III, no ID
GMFCS level IV, no ID
GMFCS level I-IV, with ID
GMFCS level V, with ID
VABS Interpersonal relationships
Age
0
10
20
30
40
50
0 2 4 6 8 10 12 14 16 18 20 22 24 26 28 30 32 34
0 2 4 6 8 10 14 18 22 26 30 34
0 2 4 6 8 10 12 14 16 18 20 22 24 26

E.

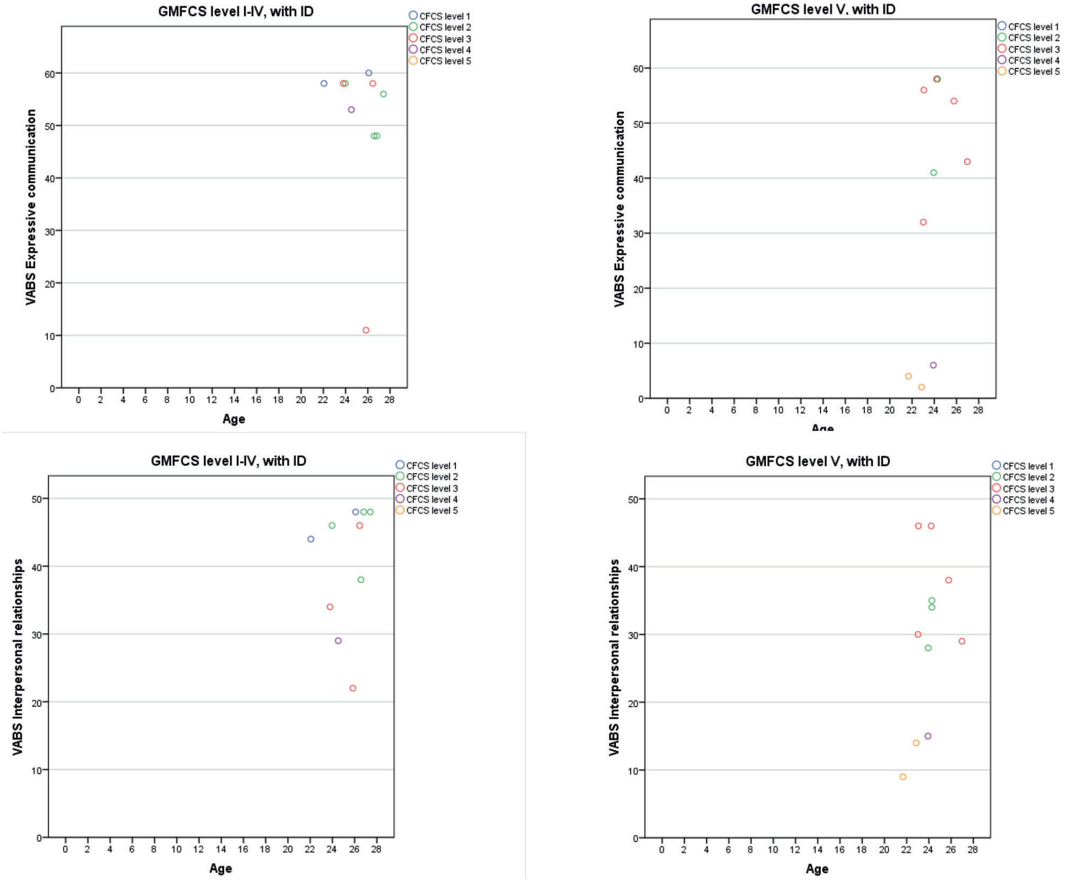

GMFCS level I, no ID
GMFCS level II, no ID
GMFCS level III, no ID
GMFCS level IV, no ID
GMFCS level I-IV, with ID
GMFCS level V, with ID
VABS Play and leisure
Age
0
10
20
30
40
0 2 4 6 8 10 12 14 16 18 20 22 24 26 28 30 32 34
0 2 4 6 8 10 12 14 16 18 20 22 24 26

F


GMFCS level I, no ID
GMFCS level II, no ID
GMFCS level III, no ID
GMFCS level IV, no ID
GMFCS level I-IV, with ID
GMFCS level V, with ID
VABS Coping skills
Age
0
10
20
30
40
0 2 4 6 8 10 12 14 16 18 20 22 24 26 28 30 32 34

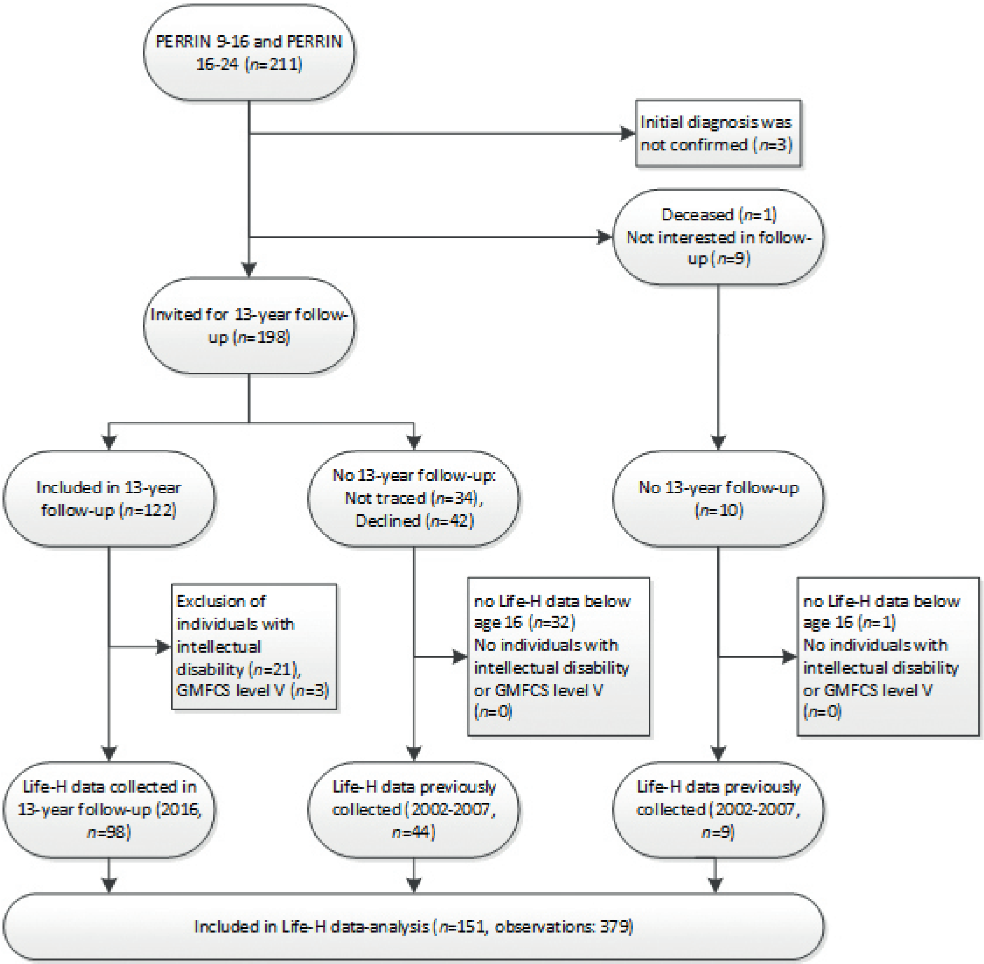
Appendix 4: Parameters of development in communication by ID and type of CP

	CP subtype	*n*	limit	(95% CI)	50% range	Age$_{90}$	(95% CI)	Residual SDs	offset
Receptive communication									
no intellectual disability	USCP	139	25.8	(25.7-25.9)	na	3y6m	(3y5m-3y8m)	1.3	0.2
	BSCP	134	25.8	(25.7-25.9)	na	3y5m	(3y4m-3y7m)	1.0	0.2
	NSCP	29	25.7	(25.5-26.0)	25.3-26.0	3y6m	(3y1m-4y0m)	0.5	0.2
intellectual disability	USCP	20	22.6	(20.3-24.9)	19.6-25.6	3y6m	(2y9m-4y7m)	2.2	0.2
	BSCP	65	20.5	(18.8-22.2)	19.6-25.6	3y3m	(2y9m-4y0m)	2.3	0.2
	NSCP	25	20.1	(16.9-23.3)	15.9-24.3	6y11m	(3y10m-12y6m)	3.1	0.2
Expressive communication									
no intellectual disability	USCP	138	61.0	(60.3-61.8)	58.9-62.0	6y2m	(5y11m-6y5m)	2.6	1.0
	BSCP	134	61.1	(60.5-61.6)	59.6-62.0	6y4m	(6y2m-6y7m)	2.5	1.0
	NSCP	29	61.5	(60.5-62.5)	60.6-62.0	7y9m	(7y2m-8y5m)	3.0	1.0
intellectual disability	USCP	20	47.2	(36.6-57.7)	36.6-57.7	7y0m	(4y8m-10y8m)	5.6	1.0
	BSCP	64	32.1	(26.1-38.2)	18.1-46.2	6y2m	(4y9m-8y3m)	4.9	1.0
	NSCP	25	29.7	(20.7-38.6)	16.3-43.1	8y5m	(4y7m-16y4m)	6.5	1.0
Written communication									
no intellectual disability	USCP	108	43.8	(42.2-45.4)	40.7- 46.0	16y0m	(15y1m-16y12m)	4.3	5.3
	BSCP	114	41.3	(39.2-43.3)	36.2-46.0	15y7m	(14y8m-16y8m)	4.1	5.3
	NSCP	28	42.5	(38.9-46.2)	38.6-46.0	15y10m	(14y8m-19y7m)	4.6	5.3
intellectual disability	USCP	11	16.1	(5.1-27.1)	6.1-26.1	14y1m	(10y7m-20y0m)	3.2	5.3
	BSCP	35	8.2	(4.0-12.3)	1.3-15.0	24y4m	(19y3m-31y3m)	2.1	5.3
	NSCP	23	5.9	(1.2-10.5)	0-12.5	22y2m	(16y9m-30y4m)	2.8	5.3

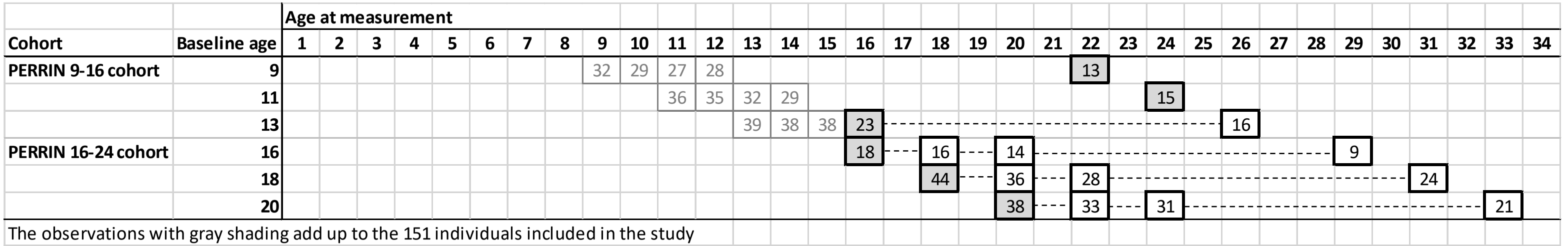


Appendix 5: Scatter plots by CFCS-level

SUPPLEMENTAL INFORMATION CHAPTER 5



Appendix 1: Flow diagram of inclusion of study participants



		Age at measurement																																	
Cohort	**Baseline age**	**1**	**2**	**3**	**4**	**5**	**6**	**7**	**8**	**9**	**10**	**11**	**12**	**13**	**14**	**15**	**16**	**17**	**18**	**19**	**20**	**21**	**22**	**23**	**24**	**25**	**26**	**27**	**28**	**29**	**30**	**31**	**32**	**33**	**34**
PERRIN 9-16 cohort	**9**									32	29	27	28										13												
	11											36	35	32	29										15										
	13													39	38	38	23										16								
PERRIN 16-24 cohort	**16**																18		16		14									9					
	18																		44		36		28									24			
	20																				38		33		31									21	

The observations with gray shading add up to the 151 individuals included in the study

Appendix 2: Number of observations at each measurement by age group (in years).

Appendix 3: Scoring Life Habits Accomplishment scale[1]

Transformed Score (0-10)	Item score (0-9)	Difficulty level	Assistance type
10	9	No difficulty	No assistance
8.89	8	No difficulty	Assistive device (or adaptation)
7.78	7	With difficulty	No assistance
6.67	6	With difficulty	Assistive device (or adaptation)
5.56	5	No difficulty	Human assistance
4.44	4	No difficulty	Assistive device (or adaptation) and human assistance
3.33	3	With difficulty	Human assistance
2.22	2	With difficulty	Assistive device (or adaptation) and human assistance
1.11	1	Accomplished by a proxy	-
0	0	Not accomplished	-

[1] Fougeyrollas P, Noreau L. *Manual: Assessment of Life Habits - General Short Form* Lac St-Charles, Québec, Canada, 2002.

Appendix 4: Scores on the Life-H for each participation domain per age interval and GMFCS level (range 0-10)

Life-H domain	GMFCS level	16-18 years			19-22 years			23-26 years			27-34 years		
		mean (SD)	<8.89	<5.56	mean (SD)	<8.89	<5.56	mean (SD)	<8.89	<5.56	mean (SD)	<8.89	<5.56
Housing	**I**	9.9 (0.5)	4%	0%	9.7 (0.6)	9%	0%	9.6 (0.8)	14%	0%	8.9 (1.1)	39%	0%
	II	9.3 (1.1)	30%	0%	9.2 (1.6)	21%	0%	8.4 (1.4)	56%	0%	7.4 (0.9)	100%	0%
	III	9.4 (0.7)	13%	0%	8.7 (0.5)	43%	0%	8.0 (1.0)	67%	0%	6.8 (1.0)	100%	0%
	IV	6.5 (1.7)	91%	27%	7.8 (1.6)	53%	7%	7.0 (1.0)	100%	13%	6.3 (1.2)	100%	25%
Interpersonal relationships	**I**	9.8 (0.4)	4%	0%	9.7 (0.6)	8%	0%	9.7 (0.5)	8%	0%	9.4 (1.2)	20%	2%
	II	9.4 (0.9)	20%	0%	9.8 (0.4)	7%	0%	9.8 (0.5)	1%	0%	9.7 (0.3)	0%	0%
	III	9.8 (0.5)	13%	0%	9.7 (0.5)	14%	0%	9.5 (0.5)	0%	0%	9.1 (0.9)	25%	0%
	IV	9.3 (0.6)	9%	0%	9.0 (1.9)	20%	7%	8.5 (1.4)	50%	0%	8.2 (1.5)	50%	0%
Education & employment	**I**	9.3 (1.3)	23%	3%	9.0 (1.6)	27%	5%	9.3 (0.8)	22%	0%	9.1 (1.2)	32%	2%
	II	8.3 (1.4)	60%	0%	8.2 (2.1)	50%	14%	9.4 (0.7)	43%	0%	7.8 (1.1)	75%	0%
	III	8.4 (1.3)	50%	0%	8.8 (1.2)	43%	0%	8.8 (1.1)	40%	0%	7.9 (1.8)	75%	25%
	IV	6.7 (2.4)	73%	36%	8.3 (1.3)	61%	8%	7.5 (3.7)	43%	14%	6.0 (1.5)	100%	38%
Recreation	**I**	9.6 (0.9)	10%	1%	9.7 (0.7)	6%	1%	9.7 (0.5)	4%	0%	9.1 (1.4)	22%	5%
	II	8.4 (2.0)	50%	10%	9.1 (1.2)	21%	0%	8.4 (1.6)	44%	11%	8.0 (2.0)	50%	0%
	III	6.7 (2.3)	62%	50%	8.5 (0.6)	71%	0%	8.5 (1.4)	50%	0%	8.1 (1.5)	50%	0%
	IV	4.9 (2.7)	91%	55%	6.6 (2.3)	80%	20%	7.6 (1.6)	62%	13%	7.8 (1.0)	62%	0%
Community life	**I**	9.6 (1.2)	8%	1%	9.8 (0.9)	2%	1%	9.9 (0.2)	0%	0%	9.7 (0.5)	5%	0%
	II	8.6 (1.7)	50%	0%	9.4 (0.8)	29%	0%	9.8 (0.4)	0%	0%	8.7 (1.0)	50%	0%
	III	7.2 (2.0)	75%	25%	9.0 (0.5)	29%	0%	9.2 (0.7)	0%	0%	8.2 (0.6)	50%	0%
	IV	5.2 (1.9)	91%	64%	6.9 (2.3)	73%	33%	8.2 (0.9)	75%	0%	8.0 (1.2)	62%	0%
Responsibilities	**I**	9.3 (1.4)	18%	3%	9.7 (0.8)	9%	0%	9.9 (0.4)	2%	0%	9.9 (0.8)	15%	0%
	II	9.6 (0.7)	20%	0%	9.7 (0.6)	7%	0%	9.5 (0.8)	11%	0%	9.9 (0.1)	0%	0%
	III	9.3 (0.8)	38%	0%	9.6 (0.7)	14%	0%	9.5 (0.5)	0%	0%	9.7 (0.6)	0%	0%
	IV	8.7 (1.8)	36%	9%	8.4 (2.3)	33%	20%	7.3 (1.9)	75%	13%	8.4 (1.6)	50%	0%

Mean (SD): mean and standard deviation of domain score Life-H
<8.89: proportions of individuals with a score below 8.89, representing participation with difficulty
<5.56: proportions of individuals with a score below 5.56, representing participation with human assistance

Appendix 5: Proportion of observations where items of Life-H were applicable, by age interval:16-18 years (no. of observations: 102), 19-22 years (no. of observations: 148), 23-26 years (no. of observations: 72) and 27-34 years (no. of observations: 57)

	% applicable	16-18 years	19-22 years	23-26 years	27-34 years
Housing	Choosing a home according to your needs	18%	51%	46%	100%
	Maintaining your home	19%	32%	65%	100%
	Maintaining the grounds or the garden	9%	4%	18%	33%
	Major household tasks	7%	21%	61%	100%
	Entering and exiting your home	98%	99%	100%	100%
	Moving around within your home	100%	100%	99%	100%
	Using the furniture	100%	100%	100%	100%
	Mobility outside your home (backyard, garden)	100%	100%	100%	100%
Interpersonal relationships	Having a sexual relationship (healthy, appropriate, safe sex)	16%	44%	53%	63%
	Maintaining an emotional relationship with your partner	19%	30%	41%	79%
	Maintaining emotional relationships with your children	0%	1%	0%	21%
	Maintaining emotional relationships with your parents	100%	100%	100%	100%
	Maintaining emotional relationships with other members of your family (brothers, sisters, uncles, etc.)	99%	99%	99%	100%
	Maintaining friendships	100%	99%	100%	96%
	Maintaining social relationships with those around you (brothers, sisters, uncles, etc.)	100%	100%	100%	100%
Employment	Choosing a career or profession	67%	80%	74%	95%
	Seeking employment	23%	37%	49%	88%
	Holding a paid job	28%	48%	71%	86%
	Using the services at your principal place of occupation (work, school, volunteer center, etc.)	85%	90%	92%	88%
	Taking part in unpaid activities (volunteering)	21%	28%	33%	49%
	Getting to, entering and getting around in your principal place of occupation (work, school, volunteer center)	92%	90%	92%	91%
	Carrying out family or home-making tasks as your main occupation	3%	1%	1%	26%

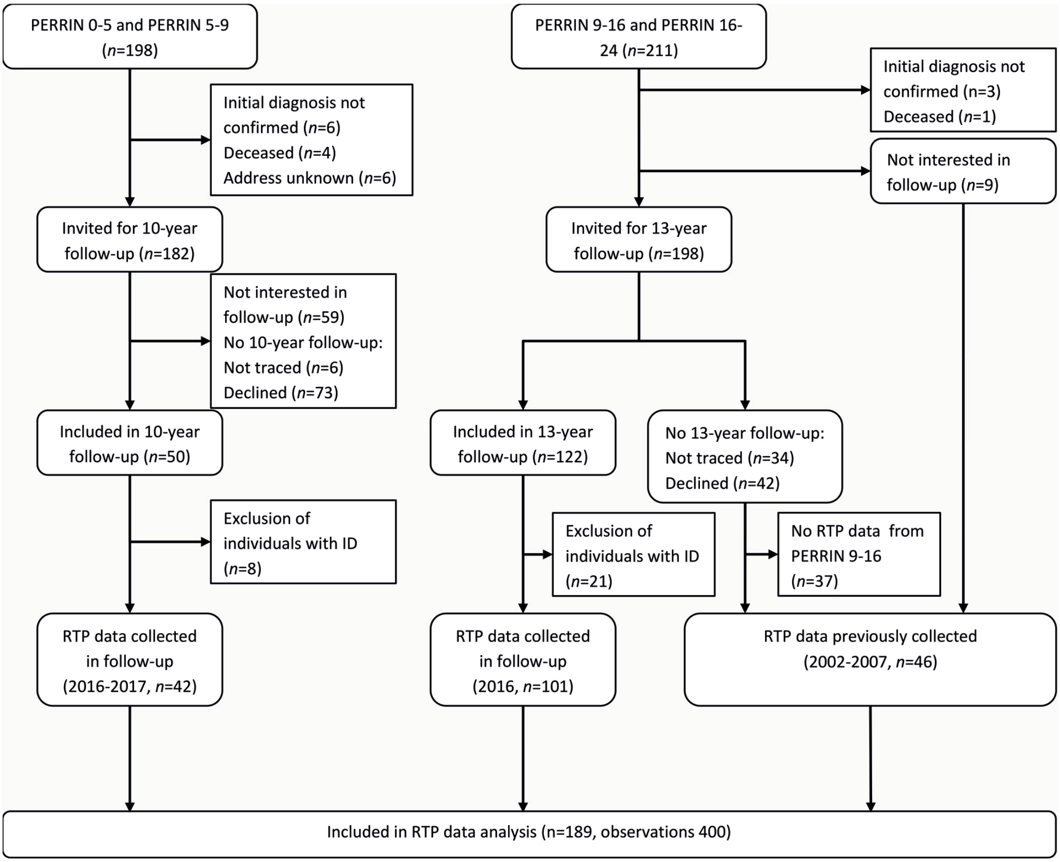
Appendix 5: Continued

	% applicable	16-18 years	19-22 years	23-26 years	27-34 years
Education	Participating in daycare or nursery school activities	2%	0%	0%	0%
	Participating in school activities or professional training at the high school level (courses, home work, extra-curricular activities)	97%	29%	8%	0%
	Undertaking professional training (trade school, university, community college)	2%	32%	43%	12%
Recreation	Participating in sporting or recreational activities (walking, sports, games, etc.)	84%	82%	83%	68%
	Participating in artistic, cultural or craft activities (music, dance, woodworking, etc.)	46%	40%	42%	39%
	Going to sporting events (hockey, baseball, etc.)	51%	47%	50%	51%
	Going to artistic or cultural events (concerts, movies, theater, etc.)	94%	96%	93%	95%
	Participating in tourist activities (traveling, visiting natural or historic sites, camping, etc.)	98%	99%	100%	96%
	Using your neighborhood recreational services (library, municipal recreation center, etc.)	77%	65%	43%	35%
Community life	Using your neighborhoods public services (health, government, banking, legal, etc.)	95%	100%	100%	100%
	Using your neighborhood businesses (supermarkets, shopping malls, dry cleaners, etc.)	100%	100%	100%	100%
	Shopping (choosing merchandise, method of payment, phone orders, etc.)	96%	99%	99%	100%
	Using eating facilities or restaurant services (table service and fast food)	97%	99%	100%	100%
	Getting to, entering and getting around inside your neighborhoods commercial buildings (supermarket, shopping malls, convenience store, etc.)	98%	100%	100%	100%
	Getting to, entering and getting around inside public buildings (health, governmental, church, recreational center, etc.)	100%	100%	100%	100%
	Participating in social or community groups (social clubs, charity or religious groups, etc.)	34%	28%	22%	30%

Appendix 5: Continued

	% applicable	16-18 years	19-22 years	23-26 years	27-34 years
Responsibilities	Recognizing the value of money and correctly using the different denominations of paper and coin money	99%	99%	100%	100%
	Planning your budget and meeting your financial obligations (spending, saving, paying bills, etc.)	70%	95%	100%	100%
	Assuming your responsibilities towards others and society (respecting the rights and property of others, voting, obeying laws, etc.)	99%	100%	100%	100%
	Assuming your personal or familial responsibilities	99%	100%	100%	100%
	Ensuring the education of your children	0%	0%	0%	10%
	Taking care of your children (health, feeding, clothing, etc.)	0%	1%	0%	21%

SUPPLEMENTAL INFORMATION CHAPTER 6



Supplementary File 1: Flowchart of participant inclusion

Supplementary File 2: The Rotterdam Transition Profile (version 2011)

Rotterdam Transition Profile

Version 2011

Information for users

Transition into adulthood is the change from one phase of life to another and is accompanied by changes in a person's environment and demands for new skills. For adolescents this means a shift towards independence and self-determination in work, housing, developing intimate relationships and financing. They take control over their life, for instance by making their own choices.
Transition into adulthood is not the same for each adolescent and is not necessarily problematic.

The Rotterdam Transition Profile presents a brief summary of the transition phases in participation domains and healthcare related domains. Completing the Transition Profile for a young person gives insight into the person's transition phase for each domain.

For transition into adulthood the following aspects are important:

a. Transition takes place in different domains of participation. Research shows that transition does not occur in all domains at the same time. Transitions will also take place in healthcare related domains.

b. Transition is a developmental process in which three phases can be distinguished:

- Phase 0. No experience at all; or phases 1 to 3 are not applicable
- Phase 1. Dependent on parents
- Phase 2. Experimenting and orientating to the future
- Phase 3. Self-reliant, autonomous

In phase 2 young persons experiment with increasing independency and self-determination. They have to develop new skills and possibly may experience problems. In this phase parents become aware that their child has to learn to make choices independently to take control over their life. Specific descriptions of the transition phases for each domain are given on the next page.

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Translators: the authors and Frederike van Markus-Doornbosch
https://www6.erasmusmc.nl/reva/onderzoek/transition/; m.roebroeck@erasmusmc.nl

Sources:

Donkervoort M, Wiegerink DJHG, van Meeteren J, Stam HJ, Roebroeck ME and the Transition Research Group South West Netherlands. Transition to adulthood: validation of the Rotterdam Transition Profile for young adults with cerebral palsy and normal intelligence. Dev Med Child Neurol 2009: 51: 53-62.

(this article) Schmidt AK, van Gorp M, van Wely L, Ketelaar M, Hilberink SR, Roebroeck ME, and the PERRIN DECADE Study Group. Autonomy in participation of individuals with cerebral palsy from youth into adulthood. … 2019

Participation domains	0	1	2	3
Education and employment **0.** Follows no education, no job **1.** General education **2.** Vocational training, work placement **3.** Paid job, volunteer work				
Finances **0.** No pocket money, fully economically dependent **1.** Pocket money, clothing allowance **2.** Job on the side, student grant **3.** Economically independent: job income, benefits				
Housing **1.** Lives with parents or caregivers, not responsible for household activities **2.** Partly responsible for household activities, domestic training, or seeking independent housing **3.** Lives independently				
Leisure (social activities) **0.** Does not arrange any leisure activities with peers **1.** Arranges leisure activities with peers at home **2.** Arranges leisure activities with peers outside the home during daytime **3.** Goes out in the evening with peers				
Intimate relationships **0.** No experience with dating **1.** Experience with dating but not yet with courtship **2.** Experience with courtship **3.** Current romantic relationship/partner				
Sexuality [a] **0.** No experience with French kissing **1.** Experience with French kissing **2.** Experience with caressing under clothes, cuddling nude **3.** Experience with sexual intercourse				
Transportation **1.** Parents or caregivers transport the adolescent/young adult **2.** Parents or caregivers arrange transportation, but they do not go with him or her **3.** Young person arranges transportation him-/herself				

Health care domains	**1**	**2**	**3**
Care demands **1.** Parents or caregivers formulate care demands **2.** Parents or caregivers, and young adult formulate demands together **3.** Young person formulates care demands him-/herself			
Services and aids **1.** Parents or caregivers apply for services and aids **2.** Young person learns the procedures to apply for services and aids **3.** Young person applies for services and aids him-/herself			
Rehabilitation services In the past year: **1.** Young person consulted paediatric rehabilitation care **2.** No consultation of rehabilitation care **3.** Young person consulted adult rehabilitation services			

[a] In the RTP version 2011 the domain Sexuality was added to discriminate between aspects of intimate relationships.

Notes regarding Schmidt et al. (2019):
In the present analyses, to ensure data consistency, the domains intimate relationships and sexuality were merged into one domain (intimate relationships), to match the validated 2007 version of the RTP (4). The study of Schmidt et al. did not include the RTP health-care domains.

Supplementary File 3: Figure observations

		Age at measurement																																	
Cohort	**Baseline age (y)**	**1**	**2**	**3**	**4**	**5**	**6**	**7**	**8**	**9**	**10**	**11**	**12**	**13**	**14**	**15**	**16**	**17**	**18**	**19**	**20**	**21**	**22**	**23**	**24**	**25**	**26**	**27**	**28**	**29**	**30**	**31**	**32**	**33**	**34**
PERRIN 0-5	**1,5**													5																					
	2,5													2																					
PERRIN 5-9	**5**															17																			
	7																	18																	
PERRIN 9-16 cohort	**9**																						13												
	11																								16										
	13																										17								
PERRIN 16-24 cohort	**16**																18[a]		16		13[a]									9					
	18																		45[a]		37[a]		27[a]									25			
	20																				38[a]		33[a]		31[a]									21	

Number of observations at each assessment presented for baseline age groups at mean age of group.

▨ Characteristics of the study population are given in table 1

[a] Missing data for leisure (baseline age 16 T_0 n=18; baseline age 18 T_0 n=45; baseline age 20 T_0 n=38, T_1 n=2, T_2 n=1), transportation (baseline age 16 T_0 n=18; baseline age 18 T_0 n=45; baseline age 20 T_0 n=38), finances (baseline age 16 T_0 n=1,T_2 n=1; baseline age 18 T_0 n=7; baseline age 20 T_0 n=2,T_2 n=1), intimate relationships (baseline age 18 T_1 n=1, T_2 n=1; baseline age 20 T_2 n=1), education and employment (baseline age 18 T_2 n=3; baseline age 20 T_2 n=1).

Supplementary File 4: Rotterdam Transition Profile characteristic

Table: Characteristic of participants

Characteristic	(n=189)
Age in years: range	12-34
Gender, n (%)	
Males	118 (62)
Females	71 (38)
Type of CP, n (%)	
Spastic unilateral	81 (43)
Spastic bilateral	85 (45)
Mixed	23 (12)
Level of gross motor function, n (%)	
GMFCS level I	130 (69)
GMFCS level II	21 (11)
GMFCS level III	16 (8)
GMFCS level IV	19 (10)
GMFCS level V	3 (2)
Level of education[a, b], n (%)	
Low	47(25)
Medium	37(20)
High	61(32)

[a] Level of education is described according to most recent information. [b] Missing data for level of education (n=44). SD, standard deviation; CP, cerebral palsy; GMFCS, Gross Motor Function Classification System.

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Movement Sciences conducts scientific research to optimize physical performance in health and disease based on a fundamental understanding of human movement in order to contribute to the fulfillment of a meaningful life.