Health Issues and Participation in Adults with Cerebral Palsy

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CONTENTS

LIST OF ABBREVIATIONS

ACSM	American College of Sports Medicine
AM	Activity monitor
BMI	Body Mass Index
BP	Blood pressure
BOSK	Dutch Association of Physically Disabled Persons and their Parents
CBS	Centraal Bureau voor de Statistiek / Statistics Netherlands
CBT	Cognitive behavioural therapy
CES-D	Center for Epidemiological Studies Depression Scale
CHART	Craig Handicap Assessment and Reporting Technique
CIQ	Community Integration Questionnaire
CP	Cerebral palsy
CVD	Cardiovascular disease
DBP	Diastolic blood pressure
ESC	European Society of Cardiology
GMFCS	Gross Motor Function Classification System
GSES-12	General Self-Efficacy Scale-12
FFM	Fat Free Mass
FIM	Functional Independence Measure
FSS	Fatigue Severity Scale
HDL-C	High-density lipoprotein cholesterol
HR	Heart rate
HRQoL	Health-related quality of life
ICF	International Classification of Functioning, Disability and Health
ISPRM	International Society of Physical and Rehabilitation Medicine
JNC 7	The seventh report of the Joint National Committee on Prevention, Detection, Evaluation and Treatment of High Blood Pressure
LIFE-H	The Assessment of Life Habits short version 3.0
MET	Metabolic equivalent
MFI-20	Multidimensional Fatigue Inventory
MORGEN	Monitoring Project on Chronic Disease Risk Factors (RIVM)
NCEP	ATPIII National Cholesterol Education Programme, Adult Treatment Panel III
PA	Physical activity
PASIPD	Physical Activity Scale for Individuals with Physical Disabilities
PMT	Psychomotor therapy
RER	Respiratory exchange ratio
ROM	Range of motion
RIVM	Rijksinstituut voor Volksgezondheid en Milieu / National Institute of Public Health and the Environment
SBP	Systolic blood pressure
SBCP	Spastic bilateral cerebral palsy
SCPE	Surveillance of Cerebral Palsy in Europe
SCORE	Systematic Coronary Risk Evaluation
SF-36	Short Form-36 Health Survey
TC	Total serum cholesterol
VAS	Visual analogue scale
VRA	Netherlands Society of Physical and Rehabilitation Medicine
WHO	World Health Organisation

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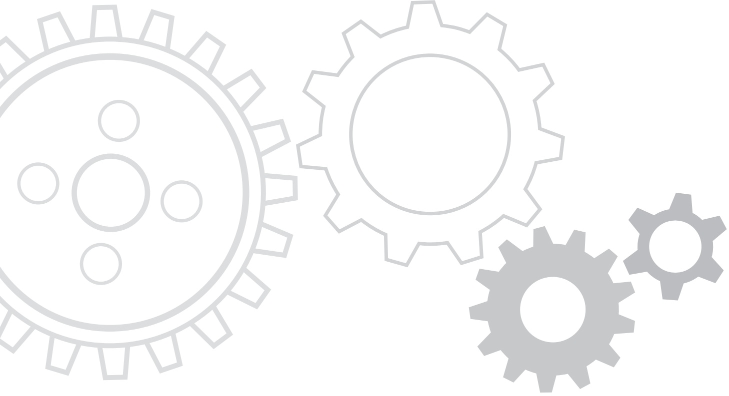
General introduction



Cerebral palsy

Brain disorders are highly prevalent, with an estimated 179 million persons affected in Europe in 2010 [1]. This thesis addresses cerebral palsy (CP), a disorder involving damage to the foetal or early infant brain. A variety of factors occurring at different times in development may lead to varying degrees of brain damage in CP. The brain lesion in itself is static. Because the brain is in charge of central controlling, damage to a developing brain may affect other organs and systems during their development. Thus, consequent body dysfunctions vary between individuals [2,3]. Therefore, CP is not considered a simple single disease entity, but as a group of disorders. The definition has been discussed and adapted over time. Nowadays, CP is regarded as a clinical descriptive term rather than an etiologic diagnosis. The current definition considers recent radiographic and neurobiologic findings: 'Cerebral palsy describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to nonprogressive disturbances that occurred in the developing foetal or infant brain. The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour, by epilepsy, and by secondary musculoskeletal problems' [3].

The definition states that abnormal motor functioning is obligatory for the diagnosis of CP. Considering the wide range of clinical presentations and degree of activity limitations, it is useful to further classify individuals with CP. For this thesis, neuromotor abnormality is classified using the Reference and Training Manual of the Surveillance of CP in Europe (SCPE) [2]. Categorisation by the activity level is according to the Gross Motor Function Classification System (GMFCS), a five-level classification system graded by the age-related severity of gross motor limitation. Distinctions between levels of motor functioning are based on functional limitations and the need for assistive devices [4,5].

Relevance of investigating persons with cerebral palsy during adulthood

CP is a common cause of disability in childhood [2,6,7], affecting 50% of Dutch paediatric rehabilitation medicine patients [8]. Prevalence ranges from 1.5 to 2.5 per 1000 live births [2,9]. In The Netherlands, the prevalence was estimated to be greater than 2 per 1000 inhabitants based on the birth years of the mid-1980s, and has been rising since the mid-1970s [10]. Although it appears that the frequency of CP amongst very low birth weight infants decreased [6], a trend towards a modest increasing prevalence was found in other Western countries as well [9-11].

This increasing prevalence may be related to improved registration and advances in neonatal and medical care [2,7,12].

It is known that persons with congenital or early acquired disabilities, such as CP, increasingly survive to older ages [13]. Nowadays, persons minimally affected by CP have life expectancies comparable to those found in the general population [14-16]. However, persons with severe CP and epilepsy have lower life expectancies [15,17].

Most children with CP receive a variety of allied health care services (e.g., physical therapy and occupational therapy). Over time, these therapies have been improved by scientific research and technological development of assistive devices. These therapies contribute to improved performance of activity and participation. However, producing significant clinical changes in impairments remains difficult. One disadvantage of relatively novel treatments, such as multilevel orthopaedic surgery for children with CP [7], is their limited availability due to treatment complexity and financial barriers, which exist even in Western countries. Thus far, no cures for CP exist.

Adults with CP tend to receive less health care than children with CP. In The Netherlands less than one-third of adults with CP receive regular care from a rehabilitation physician [18]. During their lifetimes, persons with congenital or early acquired disabilities often develop medical complications related to the natural course of their disability [13]. According to the World Health Organisation (WHO), the complications related to the primary health condition are considered secondary conditions; [19] this term is adopted in CP as well [20]. Some studies have shown that persons with congenital or early acquired disabilities have higher rates of behaviours with a health risk such as physical inactivity [19,21]. Furthermore, it is suggested that the ageing process begins as early as 40 to 50 years old [19].

The permanent nature of CP and the previously described factors suggest that adults with CP will face increasing health problems and participation restrictions, and as a consequence may consume health care resources. As shown in general populations, health problems and restricted participation may adversely affect quality of life and mood [22,23]. Considering the lifelong nature of CP, it is necessary to study each life phase. The main aim of this thesis is to provide more insight into health issues and participation amongst adults with spastic CP, in order to identify targets for prevention and treatment of health problems and methods to improve participation. With this thesis we aim to contribute to a lifespan care approach for persons with CP.

In contrast to CP in childhood, CP in adulthood is scarcely studied. At the start of the research projects described in this thesis, published literature on adults with CP was limited and various aspects of ageing with CP were scarcely understood. In daily practice we observed that adults with CP consulted rehabilitation care with various health issues that were often combined with functional loss. To reveal important research topics from a consumer's point of view, we discussed 'CP during adulthood' with the Dutch Association of Physically Disabled Persons and their Parents, BOSK [24].

These discussions and the literature available at that time gave rise to two cross-sectional studies amongst adults with spastic CP without severe cognitive impairment. The first study included persons with unilateral CP aged 25 to 35 years. In the second and larger study, we investigated adults with bilateral CP (aged 25 to 45 years), because we expected them to be at particular risk for health problems and participation restrictions. Spastic bilateral CP accounts for 50% to 60% of all cases of CP [6].

In our opinion, studying subgroups is a prerequisite to address the heterogeneity of CP. Subgroup research is also useful to facilitate comparisons with other studies, which is often restricted due to different selection criteria (e.g., enrolment based on age or CP subtype). Spastic CP was studied because it is by far the most frequent neuromotor CP subtype (about 85%) [2,6]. Persons with severe cognitive impairment [2,3] were excluded, to avoid problems associated with comprehension of study instructions and questionnaires as well as differences in life course perspectives on autonomy. We studied relatively young adults, as our studies addressed problems for which an early onset is expected. Furthermore, it is important to identify targets for preventive strategies early in life.

International Classification of Functioning, Disability and Health in rehabilitation

In Dutch rehabilitation medicine, functional status assessment and treatment are based on the WHO's 'International Classification of Functioning, Disability and Health' (ICF) [25]. Because the ICF is a framework for assessment of both individual and population levels, it is useful for scientific research as well. The ICF is used to classify health and health-related domains. These domains are classified from body, individual and societal perspectives at three levels: 1) body functions and structure, 2) activity and 3) participation. The ICF also includes a list of environmental factors and contextual information (e.g., personal factors) that affect the context in which functioning occurs. Body functions are physiological functions of body systems, including physical and psychological functions. Body structures are anatomic body parts such as organs, limbs and their components. Problems in body function or structure are impairments. Activity is the execution of a task or action by an individual. Participation is involvement in a life situation. Activity and participation are combined into one list on the ICF because extent of participation is defined based on a person's performance of a task or action in his or her current environment. The current environment brings societal context and adds 'involvement in a life situation' to participation. This thesis is subdivided into health issues and participation. The level of everyday physical activity is categorised as a health issue because of its relationship to various impairments and secondary conditions. All other activities are described in the context of participation. In Figure 1.1 the studied parameters are presented according to the ICF framework. The interaction of health and health-related domains is represented in this figure, and it illustrates the complexity of the interaction amongst impairments, activity limitations and participation restrictions in persons with CP.

Health issues

Background information

Neuromotor abnormalities in the spastic CP subtype include spasticity, muscle weakness and decreased selective motor control. The abnormal motor functioning in CP may lead to a variety of impairments, for example in movement and posture, articulation and swallowing. This thesis addresses health issues related to abnormal movement and posture.

During physical growth, symptoms like spasticity and muscle shortening may result in musculoskeletal deformities such as bony torsion, joint dislocation and muscle or tendon contracture [3,20,26]. These musculoskeletal manifestations may be accompanied by biomechanical alterations and resultant increased joint and muscle stress. The latter changes may lead to the development of secondary neuromuscular and musculoskeletal conditions, which are often aggravated by immobilisation or overuse [3,26-28].

Studies concerning secondary neuromuscular and musculoskeletal conditions in adults with CP are scarce. Murphy et al. [27] and Turk et al. [20] described a variety of conditions which reflect those encountered in our daily clinical practice. These conditions may have associated pain, deterioration of walking, and progressive functional loss [29]. Associations with fatigue, reduced fitness and physical activity are also suggested, but unclear. According to the Patient Association BOSK and our own clinical experience, these health issues may occur frequently.

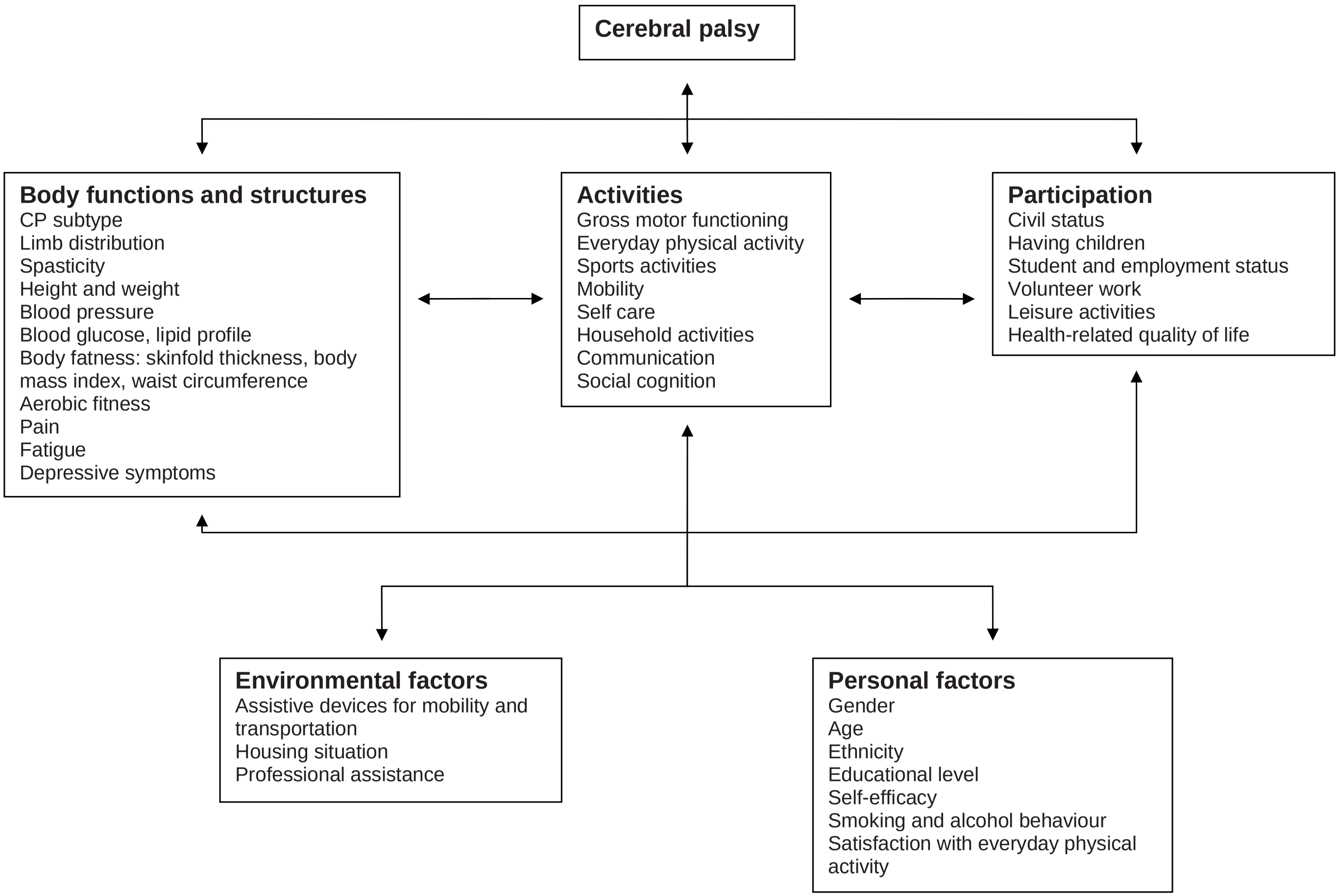


Figure 1.1 International Classification of Functioning, Disability and Health framework [25] including the parameters assessed in this thesis.

Pain and fatigue

Previous studies have shown that at least 67% of adults with CP in the USA experience chronic pain [30]. Considering the importance of pain, many later studies have addressed this issue amongst both children and adults. Pain often begins at an early age [31,32]. In the SPARCLE project, conducted with children aged 8 to 12 years, 60% had pain in the previous week, and older children reported a higher frequency of pain [32]. Recent literature shows that adults with CP have high levels of pain which frequently becomes chronic [33-35]. A Dutch study showed that adults with CP and pain more frequently consulted their rehabilitation physicians compared to adults without pain [18].

Severity and nature of fatigue have been rarely investigated in adults with CP, despite fatigue being a common symptom associated with various neurological disorders [36]. One Norwegian study demonstrated that the prevalence of fatigue was higher amongst adults with CP compared to the general population [37]. In total, 30% of adults with CP reported substantial fatigue. A recent follow-up study showed that fatigue scores were similar at 7 years' follow-up, and subgroup analysis showed that fatigue was significantly more severe in individuals with deteriorated walking [38]. Studies in general populations indicate that pain and fatigue often co-occur, especially when these symptoms are severe [39,40].

At the start of this project, data on pain and fatigue and their co-occurrence were lacking in adults with CP. We hypothesised that adults with bilateral CP have more risk to develop neuromuscular and musculoskeletal pain and fatigue compared to adults with unilateral CP.

Depressive symptoms

From a clinical and patient association point of view, depressive symptoms seemed a relevant symptom to assess. Studies amongst general populations show that pain and fatigue are associated with negative mood and depression [23,36,39,41]. However, literature is lacking on depressive symptoms and their association with pain and fatigue in adults with CP. Amongst adults with CP, a significantly higher prevalence of depression has been demonstrated amongst those without mental retardation (34.8%), as compared to those with mental retardation (12.3%) [42]. In persons with CP and chronic pain, at least 42% have depressive symptoms [43].

Aerobic fitness

In CP, the cardiorespiratory system may also be affected. The cardiorespiratory system is not fully developed prior to the brain damage which leads to CP [44]. Furthermore, it is likely that neuromotor abnormalities adversely affect the neuromotor system and exercise, which is required to maintain muscle strength and aerobic fitness [44].

Thus far, aerobic fitness has gained more attention in children with CP than in adults with CP. Children with CP show lower levels of aerobic fitness as compared to children without CP [45-48]. Prior to our research, Fernandez et al. reported that aerobic fitness was significantly lower amongst nine ambulatory adults with spastic CP (primarily bilateral) compared with healthy adults [49]. In a small sample of both ambulatory and nonambulatory adults with diverse types of CP, VO_{2peak}, as measured by using arm-crank ergometry, tended to be lower compared to that of healthy controls [50]. It is important to address aerobic fitness because of its potential to prevent secondary conditions such as pain and fatigue [21,51-53].

Everyday physical activity

The numerous health issues associated with CP [27,29,54] can restrict physical activity. Although physical activity may be beneficial to health [21], few previous studies have addressed the level of physical activity in persons with CP. Van den Berg-Emons et al. compared activity levels using the doubly-labelled water method and a respiration chamber to show that children with spastic diplegic CP are severely hypoactive [55]. Bandini et al. used doubly-labelled water and ventilation hoods to demonstrate hypoactivity in adolescent wheelchair users with CP, but not in ambulatory persons with CP [56]. Recent studies of children and adolescents with CP found similar results [57-59]. Bjornson et al. demonstrated hypoactivity using a step-watch monitor [57] and Maher et al. used self-report questionnaires to show the same [58,59]. To our knowledge, everyday physical activity [60] in adults with CP has not been objectively quantified prior to our research. Considering the subjectivity of questionnaires, it is important to measure the level of everyday physical activity objectively.

We measured the level of everyday physical activity objectively by using an accelerometry-based activity monitor (AM) [61]. The AM provides detailed information on several aspects of various mobility-related activities during daily life. We instructed participants to continue their ordinary daily life during the monitoring period, including sport and physical therapy (except swimming). According to Caspersen et al. [60] we consider sport (including exercise) not synonymous with physical activity and we regard aerobic fitness, everyday physical activity, and sport as different concepts. However, sport is a subcategory of everyday physical activity and may be performed during the AM measurement. We therefore assessed sports participation and physical therapy separately.

The low levels of aerobic fitness and physical activity demonstrated in persons with CP suggest that they may develop a cycle of deconditioning, as described by Durstine [21]: ‘disease can lead to inactivity and deconditioning. Deconditioning can lead to further inactivity and increase the potential for disability’.

According to the literature, low aerobic fitness and physical inactivity may be related to fatigue [22,36]. Although aerobic fitness, physical activity and fatigue seem to be disturbed in persons with CP, who may well benefit from physical activity or sport, studies on their interrelationship are lacking amongst adults with CP. Physical inactivity also predisposes to obesity, hypertension, type 2 diabetes and cardiovascular disease (CVD) [21,22,51].

Cardiovascular disease risk

CVD is a leading cause of morbidity and mortality worldwide and in The Netherlands [62-64]. Studies amongst general populations have demonstrated that high body fat, low aerobic fitness, and physical inactivity are risk factors for CVD [21,51,64,65]. Considering the low levels of aerobic fitness [46-50,66] and physical inactivity [55,57-59,67] found amongst persons with CP, they may be at increased risk of developing CVD [51]. Concerning body fat, conflicting results have been found in adults with CP [66]. To our knowledge, no systematic data are available on traditional CVD risk factors (e.g., hypertension, dyslipidaemia, and diabetes mellitus), or clustered CVD risk in adults with CP. It is relevant to investigate the clustered risk, because it estimates CVD risk better than the sum of individual risk factors [68].

Participation in life areas

Over time, health problems may have a detrimental effect on participation [69]. During the past decade, it has been shown that children [70-72] and young adults with CP [73] have limited participation as compared with the general population. At the start of our research, scientific knowledge on activity and participation in adults with CP was scarce. Some studies included persons of a wide range of ages, cognitive levels or CP subtypes, which limits comparison between studies. Comparison between countries is hampered by contextual factors such as accessibility to buildings, regulation, and economic factors. According to previous studies, adults with CP experience restrictions in participation in several life-areas, including employment, independent living, and social and leisure activities [29,54,74-76]. A few studies have explored whether physical complaints adversely affect participation amongst persons with CP. Turk et al. reported that pain limited activities in 56% of women with CP [20], whereas Schwartz et al. found that subjects with CP tended to report only minor interference from pain on their activity level and social or work functioning [30]. Furthermore, it has been demonstrated in persons with neurologic, orthopaedic and other medical conditions, that successful participation contributes to health and improved quality of life [77-79]. Moreover, higher participation is beneficial to society (e.g., higher levels of employment, and lower demands on social support programs). These findings support the need to study daily activities, participation and potential associated factors in adults with CP. As is specified in the ICF model [25], participation is not a direct consequence of health, but a dynamic interaction between a person and environmental factors. For a complex and lifelong disorder such as CP, exploring environmental and personal factors is also relevant. In this thesis, we focused on specific environmental factors (e.g., housing situation) and personal factors (e.g., self-efficacy), which we selected based on our clinical practice experience and Patient Association BOSK recommendations.

Health-related problems in CP are not reduced by physical interventions alone. Knowledge of personal factors may provide insight into whether psychosocial interventions can enhance behaviour, functioning and health-related quality of life (HRQoL). Self-efficacy is the belief of a person in his or her ability to organise and execute behaviours that are necessary for producing outcomes that the person desires [80]. The published literature shows evidence that self-efficacy is an important predictor of functional well-being and motivation amongst persons with disabilities, and that it is improvable by psychosocial interventions such as mentoring [81]. In young adults with disabilities, including CP, a higher level of self-efficacy is associated with participation [82].

In addition to participation outcomes, self-perceived functioning was assessed using HRQoL [83]. Health-related quality of life refers to those aspects of overall quality of life that are related to a person's physical and mental health, and functioning. In general populations, it has been shown that poor health, physical inactivity and restricted participation may negatively impact quality of life [22]. At the start of this project, data regarding HRQoL and associated factors in adults with CP were scarce. Recently, a study on HRQoL in relation to physical activity showed that Australians with CP had low levels of physical functioning and vitality [84]. Norwegian adults with CP had a lower score on most domains of HRQoL compared to the general population [85].

Aims and content of this thesis

The aim of this thesis is to provide more insight into health issues and participation amongst adults with spastic CP who do not have severe cognitive impairment. Various health issues and aspects of functioning are investigated to determine whether these are a problem in adulthood. Herein, we address frequency, severity and nature of symptoms, frequency and difficulty of participation, and consider specific environmental and personal factors. To correctly interpret results for persons with CP, we compare our findings with reference samples. To identify subgroups of persons with CP at risk, we investigate whether age, gender, GMFCS level and educational level are related to various health issues and participation. Furthermore, we explore potential associated factors. Studying associated factors of health issues and participation during adulthood may provide insight into underlying mechanisms and potential targets for improving health care for persons with CP. From a rehabilitation perspective, we are especially interested in modifiable factors. We investigate physical factors (e.g., spasticity, body fat) and exercise-oriented factors (e.g., aerobic fitness, everyday physical activity, and sport). Personal factors (e.g., self-efficacy) are studied to determine whether a psychological intervention may benefit adults with CP. Furthermore, we explore whether pain and fatigue limit participation, and which factors play a role in everyday physical activity.

Chapters 2 to 6 focus on adults with spastic bilateral CP, aged 25 to 45 years, and Chapter 7 focuses on adults with spastic unilateral CP, aged 25 to 35 years. Chapter 2 explores the prevalence and co-occurrence of chronic pain, fatigue and depressive symptoms in adults with bilateral CP. We examine associations of chronic pain and fatigue with depressive symptoms, daily activities and participation. Additionally, interrelationships amongst chronic pain, fatigue and depressive symptoms are explored. In Chapter 3 we aim to quantify objectively the level of everyday physical activity in adults with bilateral CP and compare the results with able-bodied age mates. We address whether personal and CP-related factors are associated with level of everyday physical activity, to identify subgroups at increased risk of inactivity. Chapter 4 extends on Chapters 2 and 3. It presents the level of aerobic fitness in ambulatory adults with spastic bilateral CP, and explores relationships amongst aerobic fitness, level of everyday physical activity and fatigue. Chapter 5 presents biological and lifestyle-related CVD risk factors and 10-year clustered risk of a fatal cardiovascular event in adults with bilateral CP. We explore associations between this 10-year risk and body fat, aerobic fitness and everyday physical activity, to consider modifiable exercise factors. In Chapter 6 we report on participation, HRQoL and self-efficacy in adults with spastic bilateral CP. We examine associations between participation or HRQoL, and level of education, gross motor functioning and self-efficacy. Chapter 7 investigates the level of everyday physical activity and participation amongst adults with spastic unilateral CP, as compared with able-bodied age-mates. In addition, we explore potentially associated factors of everyday physical activity: physical characteristics, cognitive and functional level, and participation and satisfaction with everyday physical activity. Against the background of these results, the strengths and limitations, clinical implications and directions for future research are discussed in Chapter 8.

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2

Chronic pain, fatigue, and depressive symptoms in adults with spastic bilateral cerebral palsy

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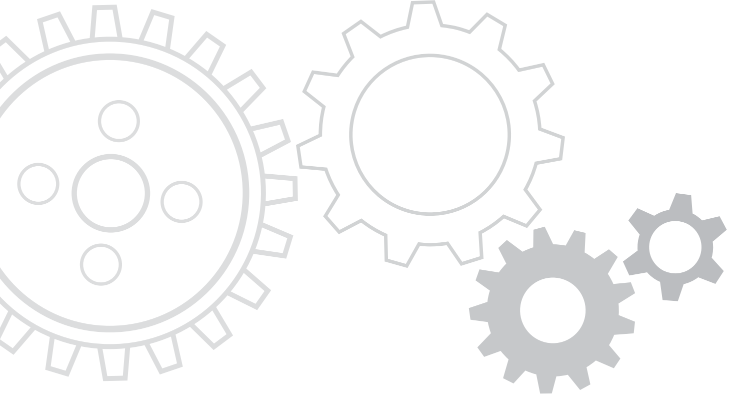

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ABSTRACT

Objective To investigate the prevalence and co-occurrence of chronic pain, fatigue and depressive symptoms in adults with spastic bilateral cerebral palsy (SBCP) and explore associations of chronic pain and fatigue with depressive symptoms and daily functioning.

Methods Fifty-six adults with SBCP without severe cognitive impairment participated (35 men, 21 women; mean age 36.4 (SD 5.8) years; Gross Motor Function Classification System level I [13], II [28], III [11], IV [4]). Chronic pain (> 3 months), severity and nature of fatigue (Fatigue Severity Scale; Multidimensional Fatigue Inventory) and depressive symptoms (Center for Epidemiological Studies Depression Scale) were assessed. Associations were explored using multivariable logistic regression analyses.

Results The study sample had a higher prevalence of chronic pain (75% versus 39%; $p < 0.001$), mean fatigue (Fatigue Severity Scale 4.4 [SD 1.3] vs 2.9 [SD 1.1]; $p < 0.001$) and prevalence of depressive symptoms (25% versus 12%; $p = 0.004$) than Dutch healthy reference samples. Chronic pain and severe fatigue co-occurred in 34% and in combination with depressive symptoms in 16% of the participants. Severity of fatigue was associated with depressive symptoms (OR = 3.38; $p < 0.01$). Chronic pain and fatigue were not associated with limitations in daily functioning.

Conclusion These findings suggest that adults with SBCP are severely affected by chronic pain, fatigue and depressive symptoms, in addition to their spastic paresis.

INTRODUCTION

Pain and fatigue are common symptoms in adults with cerebral palsy (CP) [1-4]. In CP, pain often begins at an early age and frequently becomes a chronic condition [1,2,4]. At least 67% of adults with CP in the USA experience chronic pain [4].

Fatigue is a well-known symptom in various neurological disorders, but is rarely studied in CP [3,5,6]. In a Norwegian study [3,7] the prevalence of fatigue was higher in adults with CP (30%) than in the general population (22%).

Pain and fatigue are also common symptoms in the general population [8,9] and often co-occur, especially when they are severe [8]. In the general population, pain and fatigue have been shown to impact daily activities [9] and to be associated with psychological distress, negative mood and depression [8,10]. Regarding the impact of pain and fatigue on daily activities in CP, Schwartz et al. [4] found that adults with CP tended to report only minor interference from pain on their activity level and social or work functioning, whereas Jahnsen et al. demonstrated that one-third of their CP sample reported a moderate to extreme impact of pain in daily life [2] and an association of fatigue with limitations due to physical health problems [3]. In a study among children with CP, pain and fatigue were associated with lowered school functioning and in some cases, partially explained the association between severity of CP and performance at school [5].

Very little has been reported about depression in adults with CP. Jensen et al. [11] found that in adults with CP experiencing chronic pain at least 42% had depressive symptoms and that average pain intensity was associated with depressive symptoms [12].

To our knowledge, co-occurrence of chronic pain, fatigue and depressive symptoms has not been investigated in adults with CP and objective information on these symptoms in subtypes of CP is scarce. According to Jahnsen et al., musculoskeletal pain in the back, knee, foot and ankle, as well as fatigue, tended to be more prevalent in bilateral than unilateral CP [2,3]. In our opinion, studying subgroups is a prerequisite for handling the heterogeneity of CP, therefore we focused on adults with spastic bilateral CP (SBCP) because we expected them to be particularly at risk for chronic pain and fatigue.

The present study aimed to investigate the prevalence and co-occurrence of chronic pain, fatigue and depressive symptoms in a sample of adults with SBCP without severe cognitive impairment, aged 25 to 45 years. Persons with SBCP were compared with Dutch healthy reference samples and differentiated into condition-severity subgroups using the Gross

Motor Function Classification System (GMFCS). In addition, we explored the associations of chronic pain and fatigue with depressive symptoms and daily functioning.

METHODS

Participants

This study was part of a larger study on daily functioning and physical fitness in adults with SBCP [13,14]. Participants were recruited from 10 rehabilitation centres in the western and central regions of The Netherlands and through the Association of Physically Disabled Persons and their Parents (BOSK). In addition to registers of adult rehabilitation, historical registers of paediatric rehabilitation were used to trace adults with CP, because in The Netherlands only part of the adult CP population receives rehabilitation care. The inclusion criteria were a diagnosis of SBCP and age 25 to 45 years. Exclusion criteria were any multimorbidity with lasting effects on physical activity or contraindicated for a progressive maximal ergometer test (e.g., severe cardiopulmonary disease), full dependence on electric wheelchair propulsion, inadequate Dutch language proficiency, legal inability and severe cognitive impairment according to medical files. The last criterion excluded persons who could not understand study instructions and questionnaires. Of 152 eligible persons who received the study invitation, 138 replied. Six persons were excluded because of multimorbidity. Seventy-six persons declined to participate for several reasons: a lack of time or burden of the measurement ($n = 27$), personal reasons ($n = 6$), inadequate language proficiency ($n = 2$) and no clear reason ($n = 41$). Fifty-six persons (35 men, 21 women; mean age 36.4 [SD 5.8] years) participated in the study. Characteristics of the study sample are presented in Table 2.1.

A non-response study showed that, on average, participants were older than non-participants (mean difference 2.5 years; $p < 0.01$). There was no difference in gender or distribution of limb impairment.

All participants received verbal and written information about the study and gave their written consent to participate. The Medical Ethics Committee of the Erasmus Medical Center and the participating rehabilitation centres approved the study.

Table 2.1 Characteristics of study participants

	Sample (*n* = 56)
Age, mean (SD; years)	36.4 (5.8)
Gender: men	35 (62)
Educational level [a]	
Low	15 (27)
Medium	24 (43)
High	17 (30)
GMFCS	
I	13 (23)
II	28 (50)
III	11 (20)
IV	4 (7)
Limb distribution	
Diplegia	30 (54)
Tetraplegia	26 (46)
Spasticity in most affected lower extremity [b]	
Two muscle groups	5 (10)
Three muscle groups	19 (38)
Four muscle groups	26 (52)
Life-habits: difficulty in performance	
Daily activities subdomain [c]	35 (63)
Social participation subdomain [c]	20 (36)

[a] Low: no education or elementary school and pre-vocational practical education; medium: pre-vocational theoretical education and upper secondary vocational education (vocational high school); high: secondary education, higher education, and university.
[b] Spasticity was assessed in 50 of the 56 participants.
[c] Participants reporting difficulty in daily activities or social roles (mean score < 8.0).
Values *n* (%) except where otherwise stated. GMFCS: Gross Motor Function Classification System.

Procedures

Data were collected in face-to-face interviews, which included questions on chronic pain. Self-report instruments administered in the presence of a trained researcher included: demographics, Fatigue Severity Scale (FSS) [15], Multidimensional Fatigue Inventory [16], Center for Epidemiological Studies Depression Scale (CES-D) [17,18] and Assessment of Life Habits 3.0 (LIFE-H) [19]. A physical examination was performed to assess neuromotor ability and spasticity.

Main outcome measures were chronic pain (> 3months) [20], severe fatigue (FSS ≥ 5.1) [21] and depressive symptoms (CES-D ≥ 16) [17,18]. Persons with SBCP were compared with reference values of various Dutch healthy samples: chronic pain (Musculoskeletal Complaints and Consequences Cohort study, age group 25 to 44 years, *n* = 1176) [9,22], FSS (mean age 54.2 [SD 14.8], *n* = 113) [21]; Multidimensional Fatigue Inventory (MFI-20; mean age 41.4 [SD 1.3] years, *n* = 67) [23] and CES-D (age group 25 to 44 years from a total sample of *n* = 255) [24].

Measures

Demographic and CP-related characteristics

Participants were asked to report their age, gender and level of education. Gross motor functioning was classified according to the GMFCS, which is a five-level classification system grading severity of gross motor limitations [25] and is reliable and valid in adults with CP [26]. Neuromotor abnormality [27], distribution of limb impairment (diplegia or quadriplegia) and spasticity were determined. Spasticity was assessed in four muscle groups of the lower limbs (hip adductors, hamstrings, rectus femoris, and gastrocnemius) using the Tardieu Scale [13,28]. The number of spastic muscle groups of the more affected lower limb has been reported [13].

Pain

We asked whether a person currently had pain. In participants with current pain, we assessed per localisation information on duration, frequency and possible causes. Chronic pain was defined as continuous or intermittent musculoskeletal or neuromuscular pain lasting longer than 3 months [20]. Possible causes were self-reported and defined as (1) a diagnosis made by a medical doctor or (2) pain in muscles or muscle cramps. We reported on localisation, the number of sites and possible causes of chronic pain, as well as pain lasting more than 1 year.

Fatigue

Severity of fatigue was assessed by the FSS [15]. Participants rated their agreement with nine statements concerning the severity and impact of fatigue on daily life on a scale of 1 to 7. Severe fatigue was defined as a FSS score more than 2 SDs above the mean score in healthy individuals (FSS $\geq$ 5.1) and fatigue as a score of at least 1 SD above the mean score in healthy individuals (FSS $\geq$ 4.0) [21].

The Multidimensional Fatigue Inventory is a 20-item self-report instrument designed to assess the nature of fatigue during the previous 2 weeks [16]. Scale scores range from 4 to 20, and higher scores indicate greater fatigue.

Depressive symptoms

The CES-D is a widely used self-report instrument to screen current depressive symptoms [17,18]. In the present study, the originally determined cut-off score of at least 16 (out of a maximum of 60) was used to indicate depressive symptoms based on international literature [17,18].

Daily activities and social participation

The Assessment of Life Habits 3.0 was used to assess performance in daily activities and social participation [19]. A mean score of < 8.0 (maximum = 9) on it indicated performance with difficulty [14].

More information on the questionnaires, including psychometric properties, and reference samples is presented in the supporting information in the appendix.

Statistical analysis

To compare the prevalence of chronic pain, fatigue and depressive symptoms between persons with SBCP and reference samples, one-sample *t*-tests (for means) or binomial tests (for proportions) were used. Differences between subgroups (i.e. gender, GMFCS in three subgroups [I, II and III/IV [14]]) were analysed by *t*-tests or analysis of variance (for means) and χ^2 test (for proportions). For scores with a skewed distribution, the Mann-Whitney *U* test or Kruskal-Wallis test were used. Associations of chronic pain, fatigue and depressive symptoms with demographic and CP-related characteristics were explored with univariable logistic regression analyses. For multivariable logistic regression analyses each symptom was used as a dichotomised variable (no/yes). To study fatigue as a determinant, raw sum-scores of the FSS were transformed in z-scores to allow better comparison of the strength of the determinant. Multivariable logistic regression analyses were performed to study associations of chronic pain and severe fatigue with depressive symptoms, daily activities and social participation (the latter two were dichotomised as with or without difficulty). In addition, interrelationships among chronic pain, fatigue and depressive symptoms were explored. In all these analyses we corrected for gender and GMFCS level (categorised in three subgroups: I, II, III/IV) based on clinical and scientific data [8,14]. Correcting for the distribution of limb impairment instead of GMFCS level showed comparable results. Gender and GMFCS were entered in the first block (basic model), and one of the symptoms was entered in a second block (extended model). For the multivariable models, odds ratios (OR) with 95% confidence intervals (95% CI) and *p*-values are reported. All models were validated using bootstrap analysis, generating 1000 bootstrap samples from the original data, refitting the logistic regression model to each sample, and computing the mean ORs and corresponding bias corrected 95% CI from the bootstrap estimates. Data analysis was performed using SPSS for Windows (version 16.0; SPSS, Chicago, IL, USA). The bootstrap procedure was performed with Stata 12 (StataCorp, College Station, TX, USA).

RESULTS

Chronic pain

Seventy-five percent of the adults with SBCP reported chronic pain (> 3 months) compared with 39% of the Dutch reference group in the same age range [9,22] ($p < 0.001$) (Table 2.2). Occurrence of chronic pain did not differ between men and women ($p = 0.15$) or between GMFCS level ($p = 0.47$). Chronic pain lasting more than 1 year was reported in 68% of the adults with SBCP and was more frequent in women than men (85% versus 58%, $p = 0.04$).

Localisation and the number of sites of chronic pain in adults with SBCP and the Dutch reference sample [9,22] are presented in Table 2.2. Forty-five percent of the sample described pain related to the muscles or muscle cramps and 18% reported osteoarthritis. Other self-reported causes of pain included tendinitis, arthritis, bursitis, hip dysplasia, carpal tunnel syndrome, and physical overuse.

Fatigue

Mean scores for severity of fatigue (FSS) and nature of fatigue (Multidimensional Fatigue Inventory) were higher in persons with SBCP than the Dutch reference samples [21,23] ($p < 0.01$) (Table 2.2). Twenty percent of the sample was fatigued and a further 41% were severely fatigued. Subgroup analysis showed no effects of gender ($p = 0.72$) on severity of fatigue, whereas a trend was found for GMFCS level ($p = 0.08$).

Depressive symptoms

Adults with SBCP had significantly more depressive symptoms than a Dutch reference sample aged 25 to 44 years (25% vs 12%, $p = 0.004$) (Table 2.2) [24]. No differences were found between men and women for the depressive symptoms scale ($p = 0.63$) or its subscales. Participants in

Table 2.2 Prevalence and characteristics of chronic pain, fatigue, and depressive symptoms in adults with spastic bilateral cerebral palsy (CP) and Dutch reference values

	Adults with CP (*n* = 56)		Reference samples (*n* [a])		
	n (%) or mean (SD)	Range	Mean (SD) or *n*	Reference	Difference (*p*)
Chronic pain (> 3 months)	42 (75%) (95% CI 64–86)		39% (95% CI 36–42)	9, 22	< 0.001 [b]
Pain location				22	
Neck	}18		11		
Shoulder			12		
Higher back	}39		6		
Lower back			19		
Arm	7		[c]		
Elbow	2		3		
Wrist/hand	5		7		
Leg	11		[c]		
Hip	20		4		
Knee	25		8		
Ankle	11		2		
Foot	11		3		
Whole body	4		[c]		
Number of pain sites				22	
0	25		61		
1	21		21		
2	29		}14		
3	18				
4	5		4 (≥ 4)		
5	2				
Severe fatigue (FSS ≥ 5.1)	23 (41%) (95% CI 28–54)		[e]		
Fatigue (FSS ≥ 4.0 – <5.1)	11 (20%)		[e]		
No fatigue	22 (39%)		[e]		
Mean score	4.4 (1.3)	1.8–6.8	2.9 (1.1)	21	< 0.001
Nature of fatigue (MFI-20)					
General fatigue	11.0 (4.4)	4–20	7.0 (2.9)	23	< 0.001
Physical fatigue	10.0 (4.0)	4–20	6.3 (2.4)		< 0.001
Mental fatigue	9.3 (4.4)	4–18	7.0 (2.8)		< 0.001
Reduction in activities	9.1 (3.8)	4–19	7.0 (2.8)		< 0.001
Reduction in motivation	7.7 (3.1)	4–16	6.5 (2.2)		0.005
Depressive symptoms (CES-D ≥16)	14 (25%) (95% CI 14–36)		12%	24	0.004 [b]
CES-D total score	13.4 (5.2)	6–28	7.4 (7.5)	24	< 0.001
Somatic retarded affect	3.7 (2.7)				
Depressed affect	1.4 (1.9) [d]				
Positive affect	7.4 (1.4)				
Interpersonal affect	0.5 (1.0) [d]				

[a] Numbers of reference samples: 9 and 22: *n* = 1176; 21: *n* = 113; 23: *n* = 67; 24: *n* = 255. [b] Binomial test. [c] Not assessed. [d] Skewed distribution. [e] Not available. FSS: Fatigue Severity Scale; MFI-20: Multidimensional Fatigue Inventory; CES-D: Center for Epidemiological Studies Depression scale.

GMFCS level III or IV had a higher total score, as well as higher dimension scores of somatic retarded affect and interpersonal affect ($p = 0.004$, $p = 0.013$ and $p = 0.009$, respectively) than participants in GMFCS level I or II.

Chronic pain, severe fatigue and depressive symptoms were not related to age or level of education (p-values > 0.05).

Co-occurrence and associations of chronic pain and fatigue with depressive symptoms

Results of analyses of co-occurrence of chronic pain, severe fatigue and depressive symptoms are presented in Figure 2.1. Chronic pain and severe fatigue co-occurred in 34% and in combination with depressive symptoms in 16% of the study sample. In the 42 persons with chronic pain, the prevalence of severe fatigue was 45% and depressive symptoms 29% and in those without chronic pain it was 29% and 14%, respectively. Corrected for gender and GMFCS level, chronic pain was not significantly associated with reporting severe fatigue (OR 1.76, 95% CI 0.46–6.79) or depressive symptoms (OR 1.94, 95% CI 0.35–10.64; Table 2.3).

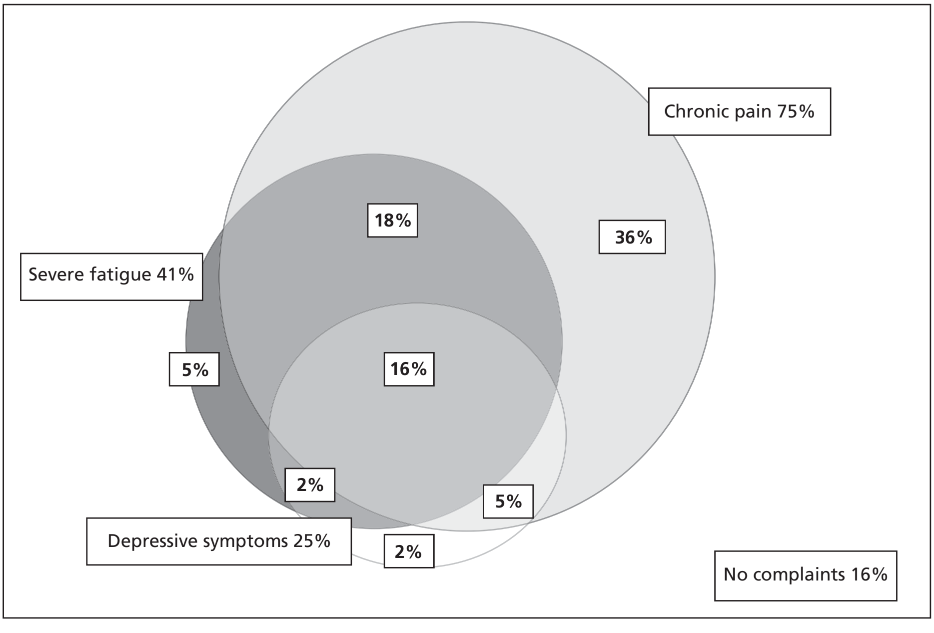

Figure 2.1 Co-occurrence of chronic pain, severe fatigue and depressive symptoms in adults with spastic bilateral cerebral palsy ($n = 56$).

Table 2.3 Interrelationships amongst chronic pain, fatigue, and depressive symptoms in adults with spastic bilateral cerebral palsy (*n* = 56)

	Severe fatigue (FSS ≥ 5.1)			Depressive symptoms (CES-D ≥ 16)		
	Basic model	Extended model	*p*	Basic model	Extended model	*p*
Independent variable	OR (95% CI)	OR (95% CI)		OR (95% CI)	OR (95% CI)	
Gender (women)	1.54 (0.51–4.68)	1.41 (0.45–4.39)		1.31 (0.36–4.72)	1.21 (0.33–4.42)	
GMFCS	1.61 (0.74–3.51)	1.55 (0.70–3.39)		2.49 (0.97–6.42)	2.38 (0.92–6.15)	
Chronic pain	[b]	1.76 (0.46–6.79)	0.41	[b]	1.94 (0.35–10.64)	0.45
	Chronic pain (> 3 months)			Depressive symptoms (CES–D ≥ 16)		
Gender (women)	2.83 (0.67–11.88)	2.67 (0.60–11.93)		1.31 (0.36–4.72)	1.01 (0.24–4.28)	
GMFCS	1.68 (0.67–4.23)	1.44 (0.53–3.95)		2.49 (0.97–6.42)	1.89 (0.72–5.00)	
Severity of fatigue (FSS) [a]	[b]	2.26 (1.08–4.72)	0.03	[b]	3.38 (1.38–8.30)	0.008

[a] Independent variable: FSS score was transformed to a z-score.

[b] Not included in regression equation. Basic model: model with gender and GMFCS level in block 1. Extended model: basic model with one of the other symptoms (listed on the third line of a model) added in block 2.

FSS: Fatigue Severity Scale; CES-D: Center for Epidemiological Studies Depression scale; GMFCS: Gross Motor Function Classification System.

In the 23 persons with severe fatigue, 83% had chronic pain and 44% depressive symptoms, compared with 70% and 12% in persons without severe fatigue. Those with more severe fatigue tended to report chronic pain (OR 2.26, 95% CI 1.08–4.72) or depressive symptoms (OR 3.38, 95% CI 1.38–8.30; Table 2.3) more often, after correcting for gender and GMFCS level. Additional bootstrap analyses showed similar results, except for no significant association between severity of fatigue and chronic pain (bootstrap OR 2.26, 95% CI 0.97–10.87).

Associations of chronic pain and fatigue with daily activities and participation

Almost two-thirds of the sample reported difficulty in performing daily activities, and one-third in social participation (Table 2.1). Chronic pain or severe fatigue was not significantly associated with difficulty in daily activities or social participation, after correcting for gender and GMFCS level (OR 0.59, 95% CI 0.15–2.39 and OR 0.86, 95% CI 0.18–3.99 for chronic pain, and OR 0.82, 95% CI 0.43–1.55 and OR 1.12, 95% CI 0.56–2.24 for more severe fatigue). These results seemed to be robust as verified by bootstrap analyses.

DISCUSSION

In this study, adults with SBCP without severe cognitive impairment, aged 25 to 45 years, showed a high prevalence of chronic pain, fatigue and depressive symptoms compared with healthy reference samples. Prevalence of these symptoms was not age- or gender specific, nor associated with GMFCS level or level of education, except for more depressive symptoms in persons with GMFCS level III or IV.

The high level of chronic pain observed in the present study is in accordance with Schwartz and et al. [4,11]. The number of pain sites was higher in SBCP than in the Dutch population of the same age [9,22]. As with previous studies in adults with CP [1,2,4], most pain sites were localised in the back, hips or lower extremities, which matches the bilateral distribution of CP. In accordance with Murphy et al. [29] , the present sample reported a variety of assumed causes of pain. These may be consequences of spasticity, musculoskeletal deformities and pathological movement patterns, possibly in combination with overuse of affected areas.

For fatigue severity, similar levels were reported by Opheim et al. [7], which emphasizes the seriousness of fatigue in adults with SBCP. Three factors may affect fatigue in this population. First, low levels of physical activity and physical fitness may play a role; in fact, in the present sample physical fitness in males was weakly associated with fatigue [2,13]. Second, for CP-related fatigue, central sensitization, psychological characteristics, or brain damage, such as observed after stroke [6], may be of influence; however their role is not yet clear. The present study showed similar levels of physical and mental fatigue in SBCP, whereas Jahnsen et al. found higher levels of physical fatigue, but not mental fatigue, in those with CP than in the general population [3,7]. Third, pain medication, anti-spastic medication, and anti-depressants may have fatigue as a side effect.

The relatively high prevalence of depressive symptoms in the present sample, especially in persons with lower gross motor functioning, suggests that their lifelong disability may coincide with feelings of depression. Other factors, such as psychological characteristics or brain damage, may also play a role, but were not studied.

Co-occurrence of chronic pain, severe fatigue and depressive symptoms seemed higher in adults with SBCP than the general Dutch population, reporting 42 to 48% chronic pain and 12% depressive symptoms in persons with fatigue and 8% depressive symptoms in persons with chronic pain [8]. The reliability of the co-occurrence rates in the present study might be limited by the small sample size.

Although associations between chronic pain and fatigue have previously been shown in adults with CP [3,4], in other diagnoses, and in the general population [8], our findings did not support this. In the bootstrap analyses the association between chronic pain and fatigue was not confirmed, which might be explained by the small sample size. Contrary to our expectations, chronic pain and depressive symptoms were not related. In addition to the small sample size, this might be due to the relatively young age of the study sample, or to their apparent acceptance of pain as part of their lifelong disability. However, other aspects of the lifelong disability, like deterioration of body functions, might contribute to the development of depressive symptoms. Severity of fatigue and depressive symptoms were associated in our study. This association has been described previously in various populations and is complex [8]. It is unlikely that it can be explained solely by the assessment methods, because fatigue (FSS) and depressive symptoms (CES-D) have been shown to be distinct entities in persons with and without chronic diseases [15]. Owing to the cross-sectional design, as well as the scarce knowledge of the complex mechanisms that might occur between the three symptoms studied, causal interpretation is limited.

In the study sample, neither chronic pain nor fatigue was associated with difficulty in daily functioning. In persons with SBCP these symptoms might not affect functioning as much as they would in able-bodied persons, because activities and participation could have been limited or adapted from an early age. In addition, persons with CP may cope with or become accustomed to pain and fatigue, which may desensitize them to any detrimental effects. Furthermore, the assessment method, which addresses the degree of difficulty and not the frequency of daily activities and participation, might have obscured any association.

Other limitations of the study include the low study participation and some aspects of the assessment methods used. First, the low study participation should be mentioned. In The Netherlands there is no national CP register, and less than one-third of the adults with CP are under regular control of a rehabilitation physician [1]. In fact, only 5% of the present study sample was currently receiving rehabilitation treatment. Consequently, adults with CP are difficult to trace back and may not be inclined to participate in research. Other reasons for the low study participation were a lack of time and burden of the research, including a 48-hour registration with accelerometry for another part of the study [13]. In comparable diagnoses, such as in former polio patients, similar problems in tracing persons and low study participation have been encountered [30]. Our non-response study identified age (mean difference of 2.5 years) as the only significantly different variable between participants and non-participants; however, because age was not associated with the symptoms studied, no bias for this parameter is expected. In addition, gender, level of education, and GMFCS level were comparable to the subgroup of adults with SBCP without severe cognitive impairment in a representative cohort from the same geographic region [31], adding to the likelihood of a representative sample.

Secondly, some sources of selection bias might have been present. Persons with fatigue or feelings of depression may have declined the study invitation. Because of the larger study's focus on physical fitness, adults with an interest in this topic may have been more likely to participate. Persons with contra-indications for a maximal ergometer test or full dependence on electric wheelchair propulsion were excluded, which might have influenced the presence of fatigue in the sample.

Finally, some aspects of the methods of assessment should be addressed. The prevalence of chronic pain, fatigue and depressive symptoms in general populations is relatively high. Therefore, to avoid overestimation of the problem of these symptoms in SBCP we compared our sample to Dutch reference samples of the healthy population. Depressive symptoms could be over-diagnosed in a population with chronic disease owing to their physical complaints. However, inclusion or exclusion of physical items of the CES-D is known to have a minimal effect on the validity of the dimension scores on this scale [10]. Moreover, physical symptoms are part of the DSM-IV diagnostic criteria of depression and should therefore be taken into account.

Despite some limitations, we believe the present results fill a gap in literature and contribute to understanding the clinical status of persons with SBCP without severe cognitive impairment at adult age. Future research should focus on developing preventive measures and more effective interventions to reduce chronic pain, fatigue and depressive symptoms in adults with SBCP.

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SUPPORTING INFORMATION ON METHODS: QUESTIONNAIRES AND REFERENCE SAMPLES

Severity of fatigue

Severity of fatigue was assessed by the Fatigue Severity Scale (FSS) [1]. Subjects rated their agreement with nine statements concerning the severity and impact of fatigue on daily life on a scale of 1 to 7. Severe fatigue was defined as a FSS score of at least two SD above the mean score in healthy individuals (FSS ≥ 5.1) and fatigue was defined as a score of at least one SD above the mean score in healthy individuals (FSS ≥ 4.0) [2]. The FSS showed high internal consistency (Cronbach's α = 0.81–0.95), high test-retest reliability (ICC = 0.86) and good validity (correlations with Visual Analogue Scales 0.47–0.81) in several groups of patients [1,2].

Nature of fatigue

The Multidimensional Fatigue Inventory (MFI-20) is a 20-item self-report instrument designed to assess the nature of fatigue during the previous 2 weeks [3]. It includes one scale assessing general fatigue and four scales assessing the nature of fatigue: physical fatigue, mental fatigue, reduction in activities, and reduction in motivation. Each scale contains four items that are rated on a five-point Likert scale, with higher scores indicating greater fatigue. Scale scores can range from 4 to 20. The psychometric properties of the MFI-20 have been studied in several groups [3-5] and it has a high internal consistency (Cronbach's α = 0.82–0.93) [3], good test-retest reliability (Pearson correlation coefficients ranging from 0.74–0.87) and satisfactory validity (correlations with Visual Analogue Scales from 0.23–0.77) [5].

Depressive symptoms

The Center for Epidemiological Studies Depression Scale (CES-D) is one of the most widely used self-report instruments to measure current depressive symptoms [6,7] The CES-D scale consists of 20 items, including four reverse-scored items, which are clustered into a total score and four dimension scores: somatic retarded affect, depressed affect, positive affect and interpersonal affect. Items are rated according to how often a subject felt that way in the previous week (0 indicates 'rarely or none of the time' and 3 indicates 'most or all of the time'). Total scores range from 0 to 60 with higher scores indicating higher levels of depressive symptoms. In the present study, the originally determined cut-off score of at least 16 was used to indicate depressive symptoms [6]. This cut-off was chosen based on international literature for the purpose of screening the study sample [7]. The CES-D has a high internal consistency (Cronbach's $\alpha = 0.85–0.94$) [6,8,9], acceptable test-retest reliability (0.45–0.70) [6,7], strong validity (high correlations with other self-report instruments, e.g. SCL-90 [$R = 0.72–0.87$]) [8] and good discriminative ability ($p < 0.01$) [8] in several populations. In past reports, the sensitivity of the CES-D for major depression varied between 60 and 99%, and its specificity varied between 73% and 94% [7,8].

Daily activities and social participation

The short version of the Assessment of Life Habits 3.0 (LIFE-H) was used to assess performance in the accomplishment of daily activities and social participation [10]. The performance score has two components: the level of difficulty when performing a life habit and the type of assistance required to perform the life habit. Both elements are combined into a scale ranging from a score of 0 for 'not accomplished or achieved', indicating total handicap, to a score of 9 for 'accomplished without difficulty', indicating optimal activity or participation. Mean scores were calculated for the sub-domains of daily activities and social participation. A mean score of less than 8.0 on the LIFE-H indicated performance with difficulty [11]. The LIFE-H has been shown to be reliable for different groups of adult patients (ICC > 0.76), also the Dutch language version; convergent validity was demonstrated by strong correlations (> 0.80) with the Impact on Participation of Autonomy Questionnaire and London Handicap Scale [12,13].

Dutch reference samples

Data on chronic pain were compared to the Dutch general population-based Musculoskeletal Complaints and Consequences Cohort study (DMC3-study; age group 25 to 44 years, $n = 1176$) [14,15]. A study by Merkies et al. [2] served as a Dutch healthy reference sample (mean age 54.2 [SD 14.8], $n = 113$) for results on severity of fatigue. The healthy sample in that study was stratified for gender and age. The sample was recruited from hospital personnel, companions (relatives, friends) of patients visiting the outpatient clinic, and volunteers unfamiliar with the study. Nature of fatigue in CP was compared with a Dutch healthy reference group (mean age 41.4 [SD 1.3] years, n = 67) 16 of unpaid, gender- and age-matched relatives of hospital employees. Results in persons with CP on depressive symptoms (CES-D) were compared with a Dutch healthy reference sample aged 25 to 44 years (of total sample of $n = 255$) [17].

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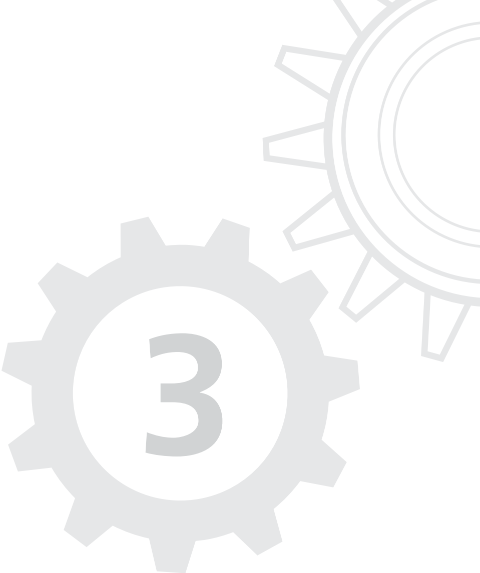

3

Inactive lifestyle in adults with spastic bilateral cerebral palsy

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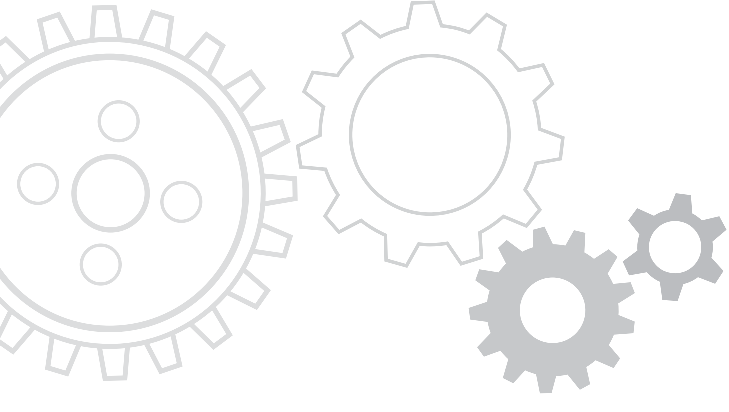

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ABSTRACT

Objective To quantify the level of everyday physical activity in adults with spastic bilateral cerebral palsy, and to study associations with personal and cerebral palsy-related characteristics.

Methods Fifty-six adults with spastic bilateral cerebral palsy (mean age 36.4 (standard deviation [SD] 5.8) years, 62% men) participated in the study. Approximately 75% had high gross motor functioning. Level of everyday physical activity was measured with an accelerometry-based Activity Monitor and was characterised by: 1) duration of dynamic activities (composite measure, percentage of 24 h); 2) intensity of activity (motility, in gravitational acceleration [g]); and 3) number of periods of continuous dynamic activity. Outcomes in adults with cerebral palsy were compared with those for able-bodied age-mates.

Results Duration of dynamic activities was 8.1 (SD 3.7) % (116 min per day), and intensity of activity was 0.020 (SD 0.007) g; both outcomes were significantly lower compared with able-bodied age-mates. Of adults with cerebral palsy, 39% had at least one period of continuous dynamic activities lasting longer than 10 min per day. Gross motor functioning was significantly associated with level of everyday physical activity (Rs = -0.34 to -0.48; $p \leq 0.01$).

Conclusion Adults with spastic bilateral cerebral palsy, especially those with low-level gross motor functioning, are at risk for an inactive lifestyle.

INTRODUCTION

Cerebral palsy (CP) is one of the most frequently occurring conditions in childhood [1]. Recently, a new definition has been developed in which CP is defined as "a group of permanent 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. The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour, by epilepsy, and by secondary musculoskeletal problems" [2]. Prevalence ranges from 1.39 to 2.80 per 1,000 live births in Europe [1]. The prevalence in The Netherlands is 1.51 per 1,000 persons, but appears to be increasing over time [3]. Life expectancy for persons with CP has increased over the past few decades and is close to that of the unaffected population for well-functioning adults with CP [4]. The most frequently occurring type of CP, the spastic form, is characterised by velocity-dependent resistance to passive movement [5].

Children with CP tend to receive much physical rehabilitation at a young age, but this attention decreases significantly with advancing age. Several studies report decreased contact with the healthcare system following completion of formal education, and care is often disrupted when disabled persons enter adulthood [6,7]. However, many persons with CP return to rehabilitation care as adults for treatment of worsening symptoms such as contractures, pain and fatigue [8,9].

This deterioration over time may lead to difficulties in performing daily activities [10] and, consequently, to an inactive lifestyle, with possible detrimental effects on physical fitness and symptoms [11]. A negative cycle may develop: inactivity leads to lower physical fitness and worsening of symptoms, which in turn lead to further inactivity. Also, comparable to persons with other disabilities, inactivity may negatively influence health-related quality of life [12] and may increase the risk of cardiovascular disease, diabetes and cancer [13].

Despite the expectation that persons with disabilities are at a high risk for an inactive lifestyle [14], only limited information is available regarding the level of everyday physical activity (PA) of adults with CP. Some evidence exists that diplegic children [15] and adolescents with certain forms of CP [16] are less physically active than able-bodied age-mates. Maher and colleagues [16] also report a strong association between level of everyday PA and gross motor functioning, with lower gross motor functioning associated with a lower level of everyday PA. These findings have been corroborated by studies of PA level in adolescents with Gross Motor

Functioning Classification System (GMFCS) level III vs those with GMFCS level I or II [17] and for non-ambulatory vs ambulatory adolescents [18]. In adults, van der Slot et al. reported no differences in level of everyday PA between persons with unilateral spastic CP and able-bodied age-matched controls [19].

To our knowledge, no objective data are available regarding level of everyday PA for adults with spastic bilateral CP. The aims of this study were, therefore: 1) to quantify the level of everyday PA for adults aged 25–45 years with spastic bilateral CP, and compare them with the levels of able-bodied age-mates; and 2) to determine whether personal and CP-related factors are associated with level of everyday PA, in order to identify subgroups at increased risk for inactivity.

METHODS

Study sample

We recruited eligible participants from 10 rehabilitation centres throughout the western and central regions of The Netherlands and via the Association of Physically Disabled Persons and Their Parents (BOSK). Inclusion criteria were a diagnosis of spastic bilateral CP (diplegia or quadriplegia) and age between 25 and 45 years. Exclusion criteria were full dependence on electric wheelchair propulsion, comorbidities impacting on PA, contraindications to progressive maximal ergometer testing (this study also evaluated maximal exercise tests for other purposes), legal inability, inadequate comprehension of the Dutch language, and cognitive impairment preventing understanding of the study protocol. An informational letter and invitation to participate was sent to eligible participants; a second letter was sent four weeks later to non-responders. All participants gave written informed consent for participation. The study was approved by the medical ethics committee of the Erasmus Medical Center and all the participating rehabilitation centres.

Measurement instruments

Personal and cerebral palsy-related characteristics

We assessed several personal and CP-related characteristics: age, gender, educational level, student/employment status, housing status, limb distribution (diplegia or quadriplegia), gross motor functioning, and spasticity.

We subdivided educational level into: 1) low, including prevocational practical education or less; 2) medium, including prevocational theoretical education and upper secondary vocational education; and 3) high, including secondary non-vocational education, higher education and university.

We classified gross motor functioning according to the GMFCS, which is based on spontaneous movements related to sitting and walking [20]. The GMFCS identifies 5 levels ranging from "walks without restrictions" (level I) to "self-mobility is severely limited even with use of assistive technology" (level V). The GMFCS was originally developed and validated for children [20], but also has demonstrated reliability and validity for describing gross motor function in adults with CP [21,22].

We assessed spasticity in 4 lower extremity muscle groups (hip adductors, hamstrings, rectus femoris and gastrocnemius) using the Tardieu Scale for clinical assessment of passive joint range of motion (ROM) [23]. The ROM for 2 different velocities was recorded, and then the difference in joint angle between these 2 measurements was calculated. The intensity of muscle reaction to stretch was scored on a scale ranging from "no resistance in whole ROM" (0) to "presence of greater than 5 cycles of clonus" (5). In a recent review, Scholtes et al. [24] concluded that the Tardieu Scale is suitable for measuring spasticity in children with CP, although it is time-consuming and lacks the standardization of muscle stretch velocity evaluation. We defined muscle spasticity as a muscle reaction intensity score of 2, 3, 4 or 5; a difference in joint angle of greater than or equal to 15°, or both. We measured spasticity bilaterally, and when differences were found, values of the most affected limb were used. The numbers of spastic muscle groups in the lower extremities on one side of the body are reported.

Level of everyday physical activity

To measure the level of everyday PA, we used an Activity Monitor (AM) (Temec Instruments BV, Kerkrade, The Netherlands). The AM is based on long-term ambulatory monitoring of signals from body-fixed accelerometers. The device consists of 4–6 accelerometers, a portable data recorder (15 x 9 x 4.5 cm; weight 700 grams) and a computer with analysis software [25]. The accelerometer signals allow calculation of movement duration, and rate and timing of activities associated with mobility (1 sec resolution). Stationary activities, such as lying, sitting and standing, can be distinguished from dynamic activities, such as walking, stair climbing, running, cycling, wheelchair propulsion (including hand-biking) and general non-cyclical movement. Furthermore, the variability of the acceleration signal (motility) can be measured as an indicator of body-segment movement intensity in which body motility addresses mean motility over a 24-h period (representing duration and intensity of everyday activity) and motility during walking and wheelchair propulsion (representing walking speed and wheelchair propulsion speed, respectively) [25, unpublished data first author]. The AM has been validated to quantify mobility-associated activities and to detect inter-group differences in levels of everyday PA [25,26].

Participants wore the AM for 48 continuous hours on randomly selected weekdays. Participants were instructed to perform their ordinary activities except they were not permitted to swim or bathe. To avoid measurement bias, we fitted AM instruments in participants' homes and explained the principles of the AM to the participants after the measurement.

For ambulatory participants, we used 4 uniaxial piezo-resistive accelerometers (Analog Devices, Breda, The Netherlands, adapted by Temec Instruments, Kerkrade, The Netherlands; size: 1.5 x 1.5 x 1 cm). We attached one accelerometer to the skin of each thigh to detect anterior-posterior direction while standing, and two accelerometers to the skin of the sternum: one to detect anterior-posterior direction, and one to detect longitudinal direction. For participants using wheelchairs, additionally to the 4 sensors that were described above, we attached one accelerometer to each wrist to detect longitudinal direction while seated with the forearm horizontal in the mid-pronation/supination position.

Accelerometers were connected to the AM and worn in padded bags around the waist. Accelerometer signals were stored digitally on a PCMCIA flash card with a 32-Hz sampling frequency. Measurements were downloaded onto a computer for kinematic analysis using Vitagraph Software. A detailed description of the activity detection procedure has been described elsewhere [25].

We measured the following data per 24-h period: 1) duration of dynamic activities as a percentage of a 24-h period (composite measure of separately detected activities of walking, wheelchair propulsion, running, cycling, and general movement); 2) number of transitions (includes all transitions except lying transitions between prone and supine positions); 3) intensity of activities: 3a) mean motility (in gravitational acceleration [g]), which reflected both duration and intensity of activity; 3b) motility during walking; 3c) motility during wheelchair propulsion; and 4) distribution of continuous dynamic activity periods (5–10 sec; 10–30 sec; 30–60 sec; 1–2 min; 2–5 min; 5–10 min; or greater than 10 min). We also computed aggregated periods of 1–5 min and greater than 5 min of continuous dynamic activities. In addition to AM measurement, we assessed participants' satisfaction with level of everyday PA using a visual analogue scale (VAS), which has demonstrated reliability and validity [27]. We asked participants to mark a 10-cm line according to their level of satisfaction with their current level of everyday PA (0 denotes "extremely dissatisfied" and 10 denotes "extremely satisfied").

Statistical analysis

Because there were no significant differences in the duration of dynamic activities between the first and second day of the measurement (paired samples t-test, $p = 0.89$), results were averaged over the 2 measurement days. Descriptive statistics were used to summarize level of everyday PA and satisfaction with level of everyday PA for the total group and for subgroups. To determine potential deficits in level of everyday PA, participant data on dynamic activity duration and mean motility were compared with those of able-bodied age-mates (age ± 5 years) ($n = 45$) using independent-samples t-tests. These age-mates were part of a large reference sample of persons without known impairments who had previously been measured with the above described AM protocol.

We examined associations between 3 main aspects of the level of everyday PA (duration of dynamic activities, mean motility and aggregate number of periods of continuous dynamic activities [1–5 min and greater than 5 min]) and personal and CP-related characteristics (age, gender, educational level, limb distribution, gross motor functioning, and spasticity), using Spearman's correlation coefficients (*R*s). When significant associations were found, we examined differences between subgroups using analysis of variance (ANOVA) with a Scheffe *post hoc*-test. Because there were few persons in GMFCS level IV and none in GMFCS level V, GMFCS levels III and IV were combined for the purpose of analysis. Statistical analyses were performed using SPSS for Windows version 12.0.1. A p-value ≤ 0.05 was considered significant.

RESULTS

Personal and cerebral palsy-related characteristics

Of 226 participants, 56 participated in the final study (response rate 25%). Reasons for refusal to participate were lack of time, lack of interest in the study, and burden to the adult with CP or caregiver. There were no differences between participants and non-participants regarding gender or affected limb distribution. On average, participants were older than non-participants (mean difference 2.5 years; t-test, $p \leq 0.01$).

The mean participant age was 36.4 (standard deviation [SD] 5.8) years; 62% were men. Affected limb was evenly distributed between quadriplegics and diplegics (Table 3.1). Most participants (73%) had high gross motor functioning (GMFCS level I or II). Seven participants used a wheelchair during the AM measurement; 3 GMFCS level IV participants used wheelchairs as their primary mode of ambulation, and 4 GMFCS level II participants

Table 3.1 Personal and cerebral palsy-related characteristics

Characteristics	Participants (*n* = 56)
Age, years, mean (SD)	36.4 (5.8)
25–29 years, *n* (%)	10 (18)
30–34 years, *n* (%)	12 (21)
35–39 years, *n* (%)	15 (27)
40–45 years, *n* (%)	19 (34)
Gender, *n* (%)	
Men	35 (62)
Women	21 (38)
Limb distribution, *n* (%)	
Diplegia	30 (54)
Quadriplegia	26 (46)
GMFCS, *n* (%) [a]	
Level I	13 (23)
Level II	28 (50)
Level III	11 (20)
Level IV	4 (7)
Level V	0 (0)
Spasticity in one lower extremity, *n* (%) [b]	
2 muscle groups	5 (10)
3 muscle groups	19 (38)
4 muscle groups	26 (52)
Educational level, *n* (%)	
High	17 (30)
Medium	24 (43)
Low	15 (27)
Student/employment, *n* (%)	
Student	2 (4)
Remunerative employment	39 (70)
Receiving social benefits	15 (26)
Housing status, *n* (%)	
Living with partner/others	16 (29)
Living alone	36 (64)
Living with parents	4 (7)

[a] Wheelchair-users were distributed over GMFCS level II (*n* = 4) and GMFCS level IV (*n* = 3).
[b] Spasticity was not assessed in 6 participants (*n* = 50).
GMFCS: Gross Motor Functioning Classification System.

used wheelchairs for long distances or participation in sports. All participants demonstrated spasticity in 2 or more muscle groups in one lower extremity. Nearly half of participants had a medium level of education and most were employed (70%). Sixty-four percent of participants lived alone, and 29% lived with a partner or others.

Level of everyday physical activity

On average, participants had a mean dynamic activity duration of 8.1 (SD 3.7) %, which corresponds to 1 h and 56 min of dynamic activities per day (Table 3.2). With regard to intensity of activities, we found a mean motility of 0.020 (SD 0.007) g. Motility during walking was 0.155 (SD 0.037) g, and motility during wheelchair propulsion was 0.034 (SD 0.011) g.

Table 3.3 shows the distribution of periods of continuous dynamic activities. Almost all participants had at least one period per day of continuous dynamic activities lasting 1–5 min. Periods of at least 5 min of continuous dynamic activity occurred for 57% of participants (range 0–6), and 39% had at least one period per day lasting longer than 10 min (range 0–2).

The mean level of satisfaction with level of everyday PA measured 6.7 (SD 2.3) cm on a scale of 0–10 cm, or moderate satisfaction (Table 3.2). Level of satisfaction was not related to the 3 main aspects of level of everyday PA (Rs = -0.13 to 0.09).

Comparison with able-bodied age-mates

In comparison with able-bodied age-mates, adults with CP had significantly shorter durations of dynamic activity (8.1% vs 10.9%, respectively; $p \leq 0.01$), and significantly lower mean motility (0.020g vs 0.027g; $p \leq 0.01$) (Figure 3.1a and b). Women with CP had significantly shorter durations of dynamic activity compared to able-bodied women (8.4% vs 12.2%, respectively; $p \leq 0.01$). For men, this difference was not statistically significant (7.8% vs 9.4%, $p = 0.11$). In both women and men, mean motility was lower compared with able-bodied age-mates (women: 0.021 g vs 0.028 g, $p = 0.03$; men: 0.019 g vs 0.027 g, $p \leq 0.01$).

Factors associated with level of everyday physical activity

Gross motor functioning was the only factor significantly associated with the 3 main aspects of level of everyday PA (Table 3.4). ANOVA and *post hoc* analyses revealed significant differences between GMFCS level I and GMFCS level III/IV participants in duration of dynamic activities

Table 3.2 Level of everyday physical activity, by level of gross motor functioning. All values are presented as means (standard deviation)

			Level of gross motor functioning					
	All ($n = 56$)	SD	GMFCS I ($n = 13$)	SD	GMFCS II ($n = 28$)	SD	GMFCS III–IV ($n = 15$)	SD
Duration of static activity (% of 24 h)	91.9	3.7	89.7 [b]	2.6	91.7	3.7	94.3	3.1
Lying	35.2	5.9	36.4	3.5	34.2	4.9	36.0	8.8
Standing	9.7	5.3	13.0	4.8	10.8	4.0	4.7	4.5
Sitting	47.1	9.4	40.3	6.7	46.7	7.2	53.6	10.9
Duration of dynamic activities (% of 24 h) [e]	8.1	3.7	10.3 [b]	2.6	8.3 [d]	3.7	5.7	3.1
General movement	2.0	1.5	2.1	1.4	2.0	1.2	2.3	2.0
Walking	5.1	3.1	7.3	1.9	5.5	2.9	2.3	2.1
Wheelchair propulsion	0.2	0.7	0	0	0.2	0.6	0.4	1.0
Cycling	0.8	1.0	0.8	0.7	0.7	1.0	0.7	1.2
Running	0	0.5	0	0.1	0	0	0	0
Mean motility (g) [a,e]	0.020	0.007	0.024 [b]	0.006	0.020 [d]	0.007	0.015	0.005
Motility during walking (g)[a]	0.155	0.037	0.168	0.030	0.160	0.035	0.133	0.042
Motility during wheelchair propulsion (g) [a]	0.034	0.011	–		0.037	0.015	0.032	0.006
Number of transitions	123	45	136	35	134	38	92	53
Periods of 1–5 min continuous dynamic activities [e]	16	11	21	7	17	13	12	9
Periods of greater than 5 min continuous dynamic activities [e]	1	1	2 [c]	2	1	2	1	1
VAS satisfaction with physical activity	6.7	2.3	6.9	2.3	6.8	2.3	6.5	2.2

[a] Mean motility and motility during walking were assessed for ambulators only ($n = 49$). Motility during wheelchair propulsion was assessed for those using a wheelchair during the measurement ($n = 7$: 4 GMFCS level II participants and 3 GMFCS level III–IV participants). Motility is expressed in g (1 g = 9.81 m/s^2).

[b] Significant difference between GMFCS level I and GMFCS level III–IV at $p \leq 0.01$.

[c] Significant difference between GMFCS level I and GMFCS level III–IV at $p \leq 0.05$.

[d] Tendency for difference between GMFCS level II and GMFCS level III–IV at $p < 0.10$.

[e] Associations with personal and CP-related characteristics were explored for these variables.

Transitions: all transitions between postures except between lying transitions. GMFCS: Gross Motor Functioning Classification System. VAS: Visual Analogue Scale.

($p \leq 0.01$), mean motility ($p \leq 0.01$), and number of periods of continuous dynamic activities greater than 5 min ($p \leq 0.05$). The difference in number of 1–5 min periods of continuous dynamic activities was not significant ($p = 0.10$) (Table 3.2).

Although not statistically significant, duration of dynamic activities and mean motility were higher in GMFCS level II compared to GMFCS level III/IV participants ($p = 0.06$ and $p = 0.09$, respectively). The number of 1 to 5 min and greater than 5 min periods of continuous

Table 3.3 Periods of continuous dynamic activities (5–10 sec; 10–30 sec; 30–60 sec; 1–2 min; 2–5 min; 5–10 min; and > 10 min)

	Number of participants with at least one period of continuous dynamic activities per day		Number of periods		
	n	%	Mean	SD	Range
5–10 sec	56	100	112	43	39–204
10–30 sec	56	100	128	53	35–279
30–60 sec	56	100	40	30	5–197
1–2 min	55	98	13	9	0–42
2–5 min	52	93	4	4	0–15
5–10 min	32	57	1	1	0–6
> 10 min	22	39	0	1	0–2

SD: standard deviation.

Table 3.4 Spearman correlations (*R*s) between personal and cerebral palsy-related characteristics and level of everyday physical activity

	Duration of dynamic activities (% of 24 h)	Mean motility (g)	Periods of continuous dynamic activities of 1–5 min (*n*)	Periods of continuous dynamic activities of > 5 min (*n*)
Age	0.00	-0.11	-0.04	-0.08
Gender	0.04	0.09	0.06	-0.24
Level of education	-0.12	-0.09	-0.18	-0.08
Limb distribution	-0.24	-0.25	-0.11	0.02
GMFCS	-0.48*	-0.46*	-0.37*	-0.34*
Spasticity	0.01	0.09	0.08	0.15

* Significant association at $p \leq 0.01$.
GMFCS: Gross Motor Functioning Classification System.

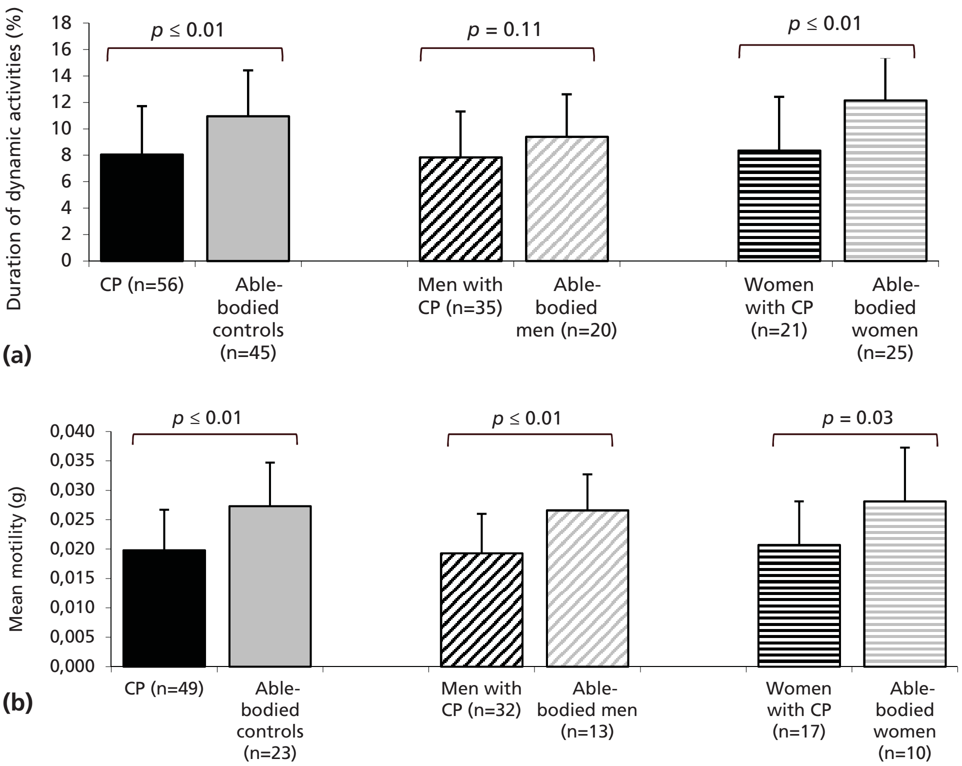

Figure 3.1 **(a)** Duration of dynamic activities in adults with spastic bilateral cerebral palsy and able-bodied age-mates, as percentage of a 24-h period. **(b)** Intensity of activities (mean motility) in adults with spastic bilateral cerebral palsy and in able-bodied age-mates, expressed in gravitational acceleration (1 g = 9.81 m/s^2).

dynamic activities did not differ between GMFCS level II and GMFCS level III/IV participants ($p = 0.32$ and $p = 0.58$). In addition, differences between GMFCS level I and GMFCS II were not significant for the three main aspects of level of everyday PA.

DISCUSSION

This is the first study objectively measuring level of everyday PA in adults with spastic bilateral CP. We have demonstrated that adults with spastic bilateral CP, and particularly those with low-level gross motor functioning, have inactive lifestyles when compared with able-bodied age-mates. Personal and CP-related characteristics other than gross motor functioning were not associated with level of everyday PA.

Level of everyday physical activity in adults with cerebral palsy

On average, participants were dynamically active for nearly 2 h per day (116 min), which is higher than levels reported in a study of adults aged 19–66 years, who were only active an average of 52 min per day [28]. However, this latter study used self-report questionnaires, which may be susceptible to social desirability and recall bias [29] and which may not capture all activities of daily living that are challenging for disabled persons [30]. A previous study using the same measurement procedures as the current study showed that ambulatory adults with spastic unilateral CP aged 25–35 years were dynamically active for 152 min per day, a finding that did not differ significantly from able-bodied age-mates [19]. These results were predictable given that study participants had near-normal lower extremity muscle tone and therefore mobility-related activities were not likely to be limited [19].

In contrast to findings in adults with unilateral CP [19], adults with bilateral CP in our study were significantly less physically active than able-bodied age-mates (excluding differences in mean duration of dynamic activities for men with CP and able-bodied age-mates, which were not significant). The inactive lifestyle reported in this study is consistent with studies of diplegic children [15] and adolescents [16-18] in which different measurement procedures, including doubly-labelled water, step counts and questionnaires, demonstrated lower levels of everyday PA in persons with CP compared with able-bodied age-mates.

In the present study, only 39% of participants had one or two periods of continuous dynamic activities for at least 10 min per day. Furthermore, only 57% had at least one period of continuous dynamic activity lasting 5–10 min each day. To maintain and promote health, able-bodied persons are recommended to engage in 30 min of moderate intensity PA each day, which can be divided into 10-min intervals [31]. Most adults with CP in our study did not achieve this minimum recommended level. This failure to achieve PA goals has also been demonstrated in other studies of adults with CP [28], and other physically disabled persons [32]. It is uncertain whether guidelines for the general population are appropriate for persons with disabilities such as CP [14]. Also, the intensity of activities performed by study participants is unknown. We suspect that adults with CP experience greater physical strain compared with the general population for similar activities; however, it is unknown if this level of exertion meets the moderate intensity exercise goals. Further research concerning the level of physical strain during everyday PA is needed.

Low levels of everyday PA in adults with CP may be explained by higher energy requirements for daily activities due to reduced muscle mass or inefficient locomotion [30]. Increased energy expenditure during everyday physical activities such as walking has been reported in children [33] and adults with CP [34]. Published data also supports a relationship between level of everyday PA and energy expended during walking [35]. Factors such as a lower physical fitness, fatigue, and pain may contribute to lower levels of everyday PA. Another reason for lower activity levels could be limited opportunities for activities such as sports participation. Several barriers exist for disabled persons, including transportation, access to equipment and facilities, and lack of awareness of facilities [36].

We found no association between mean duration of dynamic activities and satisfaction with level of everyday PA. This is in contrast with findings of van der Slot and colleagues [19], where adults with spastic unilateral CP with longer durations of dynamic activities were less satisfied with level of everyday PA. Adults with unilateral involvement may function at a higher level of everyday PA, and may therefore perform more physically demanding tasks or set higher personal goals (and have higher expectations) for functioning. The adults in our study with bilateral involvement did not attain levels of dynamic activities as high as the participants of the aforementioned study and since they might not be capable of higher levels of PA they may be quite satisfied with their current level of everyday PA.

Factors associated with level of everyday physical activity

Only gross motor functioning was associated with level of everyday PA in the present study. Inactive lifestyles were mostly found in adults with low-level gross motor functioning (GMFCS level III–IV); this finding is consistent with previous studies in adolescents with CP [16-18]. Evaluating our findings in the context of the published literature is difficult because other studies of adults with CP use different definitions and measurement methods [37,38], or do not report GMFCS level [19,28,38]. Previous studies provide contradictory data regarding level of everyday PA; some authors report no relationship [19,38] whereas others report that lower levels of motor functioning are associated with less PA [28,37].

In our study, level of everyday PA for persons with the high-level gross motor function (GMFCS level I; mean duration of dynamic activities 10.3%) was similar to that of able-bodied age-mates (10.9%). Similar results have been found in adolescents with CP [17] and, although not reported in their publication, in the study of van der Slot et al. among adults with spastic unilateral CP [19]. In the latter sample, ambulatory adults with GMFCS level I had a mean duration of dynamic activities of 11.1%, compared with 11.2% of able-bodied age-mates (personal communication).

We did not find associations between other personal and CP-related characteristics and level of everyday PA. The lack of a relationship with age and educational level is consistent with findings in adults with unilateral CP [19], but may also be explained by homogeneity of personal characteristics in the current study. Other studies in adolescents [16,17,36] and adults [19] did not report gender differences in level of everyday PA. To our knowledge, no information is available regarding the relationship between level of everyday PA and spasticity or affected limb distribution.

Limitations of the study

There are some noteworthy limitations of using the AM to measure level of everyday PA. First, we measured level of everyday PA over 2 days, but it has been suggested that at least 3–5 days of monitoring may be necessary to characterize habitual PA patterns [39]. Secondly, we may have underestimated the level of everyday PA because subjects could not swim during measurements. In several studies, swimming is noted as a frequent PA of persons with CP [37,38]. The size and the wires of the AM may also have hampered some activities in daily life. We asked the participants whether they performed their regular activities during the measurement period and this was confirmed, so influence of the measurement equipment itself is considered to be little. Additionally, we focused on comparing level of everyday PA to that of able-bodied age-mates; because able-bodied age-mates also were not permitted to swim and were also possibly hampered by the measurement equipment, these factors are believed not to have an effect on study results. Fourthly, we may have overestimated the level of everyday PA because of selection bias. Adults with CP who are interested in fitness (and may therefore have had a higher level of everyday PA) may have been more likely to participate than those with less interest.

Implications for treatment

Our study shows that adults with spastic bilateral CP have inactive lifestyles compared with able-bodied age-mates, and that they fail to achieve recommended activity levels. Because particularly adults with low-level gross motor functioning (GMFCS level III or IV) have a low level of everyday PA, interventions to increase the level of everyday PA should be targeted to this group. However, even participants with GMFCS level I and II (who achieved mean duration of dynamic activities and mean motility levels comparable to able-bodied age-mates) did not achieve minimum exercise recommendations. Furthermore, we expect that disabled persons may experience increased physical complaints secondary to ageing and functional deterioration [14,40], which may lead to reduced levels of everyday PA. We therefore believe that adults with a relatively high-level gross motor functioning would also benefit from lifestyles that are more active. Studies into the relationships between level of everyday PA, health-related fitness, and fatigue and pain symptoms may further elucidate the significance of PA to optimize health in adults with CP.

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4

Physical fitness, everyday physical activity, and fatigue in ambulatory adults with spastic bilateral cerebral palsy

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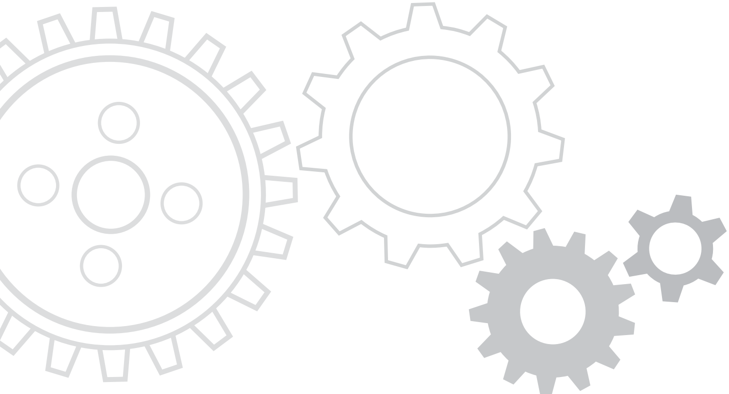

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ABSTRACT

Objective This study assessed physical fitness and its relationships with everyday physical activity (PA) and fatigue in cerebral palsy (CP).

Methods Participants were 42 adults with ambulatory spastic bilateral CP (mean age 36.4 [SD 5.8] years); 69% men; 81% with good gross motor functioning). Progressive maximal aerobic cycle tests determined VO_{2peak} (L/min). Objective levels of everyday PA were measured with accelerometry and self-reported levels of everyday PA with the Physical Activity Scale for Individuals with Physical Disabilities. Fatigue was assessed with the Fatigue Severity Scale.

Results The average aerobic capacity of adults with CP was 77% of Dutch reference values. Participants were physically active during 124 min/day (85% of Dutch reference values), and half experienced fatigue. In women, lower physical fitness was related to lower self-reported levels of PA ($R_p = 0.61$, $p = 0.03$) and in men to higher levels of fatigue ($R_p = -0.37$, $p = 0.05$). Other relationships were not significant.

Conclusion Results suggest that ambulatory adults with CP have low levels of physical fitness, are less physically active than able-bodied age mates and often experience fatigue. We found little evidence for relationships between the level of physical fitness and everyday PA or fatigue.

INTRODUCTION

Despite high survival rates for adults with cerebral palsy (CP) nowadays [1,2], consequences of CP in adulthood are often poorly understood [3]. With ageing, health issues such as the risk for cardiovascular disease and age-related decline in function are increasingly important to consider. In order to improve health throughout the life span for persons with CP, more attention toward a healthy lifestyle appears warranted.

In the general population, physical fitness is considered of major benefit to a healthy lifestyle [4,5] and is related to the prevalence of cardiovascular disease and diabetes mellitus [6-8]. In persons with CP, physical fitness may be even more important to offset the decline in function that might occur with ageing and deterioration of CP-related impairments such as reduced range of motion and increased spasticity or pain [5]. For example, Opheim et al. [9] found that the deterioration in walking function in adults with CP was associated with fatigue, and the authors attributed this fatigue presumably to low physical fitness. It has been hypothesised that fatigue may be the consequence of an imbalance between physical load capacity and physical load [10,11]. Physical load capacity may be reflected by the level of physical fitness, and physical load may be reflected by the level of everyday physical activity (PA).

Little is known about physical fitness in adults with CP, because research has focused almost entirely on children and adolescents [5]. In a group of nine ambulatory adults with spastic CP (primarily bilateral), Fernandez et al. [12] reported levels of physical fitness (VO_{2peak}) that were significantly lower than the reference values of able-bodied adults. By using arm-crank ergometry tests, Tobimatsu et al. [13] reported in both ambulatory (n = 5) and nonambulatory (n = 7) adults with diverse types of CP (spastic, ataxic, athetoid) that the VO_{2peak} tended to be lower than in able-bodied controls.

With regard to level of everyday PA in adults with CP, we found in a previous study [14], using accelerometry-based activity monitoring, that adults with spastic bilateral CP were less physically active than able-bodied age mates; however, adults with unilateral CP were equally as active as controls [15]. From studies in Norway in hemiplegic and diplegic adults with CP, we know that fatigue is a common problem in CP; 30% of participants experienced substantial fatigue, and 17% reported chronic fatigue, mainly those with bilateral CP [9,10,16].

To our knowledge, the level of physical fitness and its relationships with the level of everyday PA and fatigue have not been systematically studied yet in adults with CP. The aim of this study was, therefore, to assess the level of physical fitness and its relationships with the level of everyday PA and fatigue in adults with spastic bilateral CP. In this study, we focused on adults with diplegia or quadriplegia because we expected them to be particularly at risk for inactivity and impairments such as walking deterioration and fatigue.

METHODS

Study sample

We collaborated with 10 rehabilitation centres and rehabilitation departments from university hospitals in the western and central regions of The Netherlands and with the Association of Physically Disabled Persons and their Parents (BOSK) to include participants for the current study. Inclusion criteria were a diagnosis of spastic bilateral CP (diplegia or quadriplegia), being ambulatory with or without walking aids in daily life (Gross Motor Functioning Classification System [GMFCS] level I–III [17]), and age between 25 and 45 years. Exclusion criteria were comorbidities impacting on PA, cardiovascular contraindications to maximal ergometer testing (such as severe heart disease), legal inability, inadequate comprehension of the Dutch language, and cognitive impairment preventing understanding of the study protocol.

This study is part of a larger study [14] focusing on the level of everyday PA in 56 adults with CP. From this original cohort, four persons were nonambulatory, leaving 52 eligible participants for this part of the study. In total, 10 of these persons had missing values for aerobic capacity, either because of logistical problems, technical problems, impaired balance or knee problems. Thus, the final sample for the current study consisted of 42 adults with spastic bilateral CP. All participants gave written informed consent to participate. This study was approved by the Medical Ethics Committee of the Erasmus Medical Center and by the medical ethical committees of all participating rehabilitation centres and hospitals.

Measurements

Personal and cerebral palsy-related characteristics

We determined the following personal and CP-related characteristics: age, gender, body mass, height, level of education, limb distribution of paresis (diplegia or quadriplegia), and level of gross motor functioning and spasticity. Educational level was subdivided into: 1) low, including prevocational practical education or less; 2) medium, including prevocational theoretical education and upper secondary vocational education; and 3) high, including secondary nonvocational education, higher education, and university.

The level of gross motor functioning was assessed with the GMFCS [17]. The number of spastic muscle groups in the lower extremity on one side of the body is reported, assessed in four muscle groups of the lower extremities (hip adductors, hamstrings, rectus femoris, and gastrocnemius) with the Tardieu scale [14,18]. Spasticity was defined as a muscle reaction intensity score of 2, 3, 4, or 5 or a difference in joint angle of $\geq 15°$, or both [14,18].

Physical fitness

Aerobic capacity was used as an indicator for physical fitness. Participants performed a progressive maximal aerobic test on an electronically braked cycle ergometer (Jaeger ER 800; Jaeger Toennies, Breda, The Netherlands), according to the McMaster All-Out Progressive Continuous Protocol [19]. In addition to a screening of contraindications by a physician, all participants filled in a checklist according to the Physical Activity Readiness Questionnaire [20] before testing, to assure that there were no contraindications for a progressive maximal aerobic test. During the maximal aerobic test, a physician was on call.

The test began with a 3-min warm-up period at a low resistance (20 W), which was followed by a 5-min period of rest. According to the cycling ability of the participant, every 2 minutes the resistance was increased at a variable rate following the McMaster All-Out Progressive Continuous Protocol [19]. Both the amount of the initial resistance and the magnitude of the increase in resistance varied according to the cycling ability of the participant with increases in resistance becoming gradually smaller as the test proceeded. The pedal rate was between 40 and 60 rpm, and we gave strong verbal encouragement throughout the test. The test was terminated when the participant voluntarily stopped because of exhaustion or was unable to maintain a pedal rate of > 40 rpm.

Gas exchange and heart rate (HR) were measured continuously by using a breath-by-breath portable measurement system (K4b2; Cosmed, Rome, Italy). We calibrated the system before each test with reference gasses. We measured the HR with a transmitter belt around the chest (Polar Electro, Kempele, Finland). As the reference standard for aerobic capacity, we used the mean oxygen uptake during the last 30 seconds of exercise (VO_{2peak}, expressed in L/min). In addition, we compared VO_{2peak} (L/min) of participants with reference values of Dutch sedentary men and women in comparable age groups [21] and expressed the VO_{2peak} as a percentage of these reference values. We defined peak power output (W) as the highest work load obtained during the test.

As objective criteria for maximal exercise, we used the peak HR as a percentage of predicted HR (220 – age; at or above 95%) and respiratory exchange ratio (RER) of > 1.10. We also measured perceived strain directly after termination of the test with the modified Borg Category Scale for Rating of Perceived Exertion [22]; participants indicated how strenuous they had found the test by giving a score from 0 (no effort at all) to 10 (maximal effort).

Level of everyday physical activity

Objective level of everyday physical activity

We assessed objective PA by using an activity monitor (AM; Temec Instruments, Kerkrade, The Netherlands), which conducts long-term ambulatory monitoring of signals from four body-fixed accelerometers. The AM determines duration, rate, and moment of occurrence of postures and dynamic activities [23]. The AM has been validated to quantify mobility-associated activities and to detect intergroup differences in levels of everyday PA [23]. Participants wore the AM for 48 continuous hours on randomly selected weekdays, and they were instructed to perform their ordinary activities, except for swimming and bathing. To avoid measurement bias, we fitted AM instruments during the participants' daily life at home or work and explained the principles of the AM only after the measurement.

We calculated the duration of dynamic activities as a percentage of a 24-h period (the composite measure of separately detected activities of walking [including stair climbing and running], cycling, and general noncyclic movement) and expressed the duration of dynamic activities as a percentage of the reference values of able-bodied age mates (n = 45, measured with the same measurement protocol). The activity detection procedure [23] and other characteristics of objective PA in the same study cohort are published elsewhere [14].

Self-reported level of everyday physical activity

We determined self-reported PA by using the 7-day recall Physical Activity Scale for Individuals with Physical Disabilities (PASIPD) [24]. We used the Dutch version, which consists of 12 items concerning leisure time, household activities, and work-related physical activities. Participants were asked to recall the number of days in the past 7 days in which they participated in the activities. They also indicated how many hours per day they participated, on average, in these activities. We calculated total PASIPD scores according to Washburn et al. [24]; for each item, we multiplied the average hours spent per day by a metabolic equivalent (MET) value associated with the intensity of activity. The sum of these scores is the total

PASIPD score. The PASIPD has a test-retest reliability and criterion validity comparable with well-established, self-report PA questionnaires from the general population [25].

Fatigue

Severity of fatigue was assessed under the supervision of the researcher, with the Dutch version of the Fatigue Severity Scale (FSS) [26]. The FSS is a nine-item, self-administered questionnaire with scores ranging from 1 (strongly disagree) to 7 (strongly agree). The mean score of the nine items ranges from 1 (no signs of fatigue) to 7 (most disabling fatigue). In the current study, we defined severe fatigue as a score on the FSS of more than or equal to two standard deviations (SD) above the mean for healthy individuals (FSS ≥ 5.1) and fatigue as a score of > 1 SD above the mean (4.0 ≤ FSS score < 5.1) [27]. Internal consistency, reliability, validity, and sensitivity of the FSS have been established in several patient groups [26,27].

Statistical analysis

We used descriptive statistics to describe physical fitness, level of everyday PA, and fatigue. Because there were no significant differences in the duration of dynamic activities between the first and second days of the measurement (paired samples *t*-test, 8.7 [SD 3.7]% vs 8.5 [SD 3.7]%, $p = 0.53$), results of objective PA were averaged over the 2 measurement days. Differences between men and women and between participants and reference values were tested with independent samples *t*-tests. Relationships between physical fitness (VO_{2peak}), objective PA (duration of dynamic activities), self-reported PA (total PASIPD score), and fatigue (mean FSS score) were assessed with Pearson correlation coefficients (R_p) and were reported for men and women separately. We performed statistical analyses using SPSS for Windows version 16.0. We considered a *p*-value of ≤ 0.05 to indicate significance.

RESULTS

Study sample

Forty-two adults with spastic bilateral CP with a mean age of 36.4 (SD 5.8) years participated (Table 4.1). Limb distribution was evenly distributed between diplegia and quadriplegia, and 88% of the participants had three or four spastic muscle groups in a lower extremity. The majority had a good level of gross motor functioning (81% in GMFCS level I or II) and a medium to high level of education (76%).

Table 4.1 Personal and cerebral palsy–related characteristics

	Participants (*n* = 42)	Men (*n* = 29)	Women (*n* = 13)
Age (years), mean (SD)	36.4 (5.8)	36.7 (5.4)	35.6 (6.9)
Height (cm), mean (SD)	174.7 (8.4)	178.4 (6.8)	166.3 (4.7)
Body mass (kg), mean (SD)	70.5 (14.9)	74.3 (15.7)	61.9 (8.0)
Limb distribution, *n* (%)			
Diplegia	22 (52)	14 (48)	8 (61)
Quadriplegia	20 (48)	15 (52)	5 (39)
Gross motor functioning, *n* (%)			
GMFCS level I	11 (26)	6 (21)	5 (39)
GMFCS level II	23 (55)	18 (62)	5 (39)
GMFCS level III	8 (19)	5 (17)	3 (22)
Spastic in lower extremity, *n* (%) [a]			
2 muscle groups	4 (10)	4 (14)	0 (0)
3 muscle groups	15 (37)	11 (39)	4 (31)
4 muscle groups	22 (53)	13 (47)	9 (69)
Level of education, *n* (%)			
High	12 (28)	7 (24)	5 (39)
Medium	20 (48)	14 (48)	6 (46)
Low	10 (24)	8 (28)	2 (15)

[a] Spasticity was not assessed in one male participant, *n* = 41 (total); *n* = 28 (men).
GMFCS: Gross Motor Functioning Classification System.

Physical fitness

According to objective and subjective criteria, participants, on average, reached the maximal exercise performance. The mean peak HR was 175 (SD 17) beats/min, which resembles 96 (SD 9)% of the age-predicted maximum HR values, and the mean RER was 1.19 (SD 0.12). The mean score of perceived exertion was 9.00 (SD 1.68), indicating that participants experienced the exercise as being very heavy to maximal effort.

Table 4.2 describes physical fitness for the total group and for gender subgroups. Average aerobic capacity, defined as VO_{2peak}, was 2.40 (SD 0.39) L/min for men and 1.73 (SD 0.32) L/min for women, which was significantly lower than age-based and gender-based reference values for sedentary Dutch men (3.18 [SD 0.25] L/min, $p = 0.03$) and women (2.15 [SD 0.30] L/min, $p < 0.01$). Men had higher levels of physical fitness than women, also after correction for body mass ($p < 0.01$).

Table 4.2 Physical fitness, level of everyday physical activity, and fatigue in adults with cerebral palsy

	Total ($n = 42$)	Men ($n = 29$)	Women ($n = 13$)	p-value *
Physical fitness				
VO_{2peak} (L/min)	2.19 (0.48)	2.40 (0.39)	1.73 (0.32)	< 0.001 [a]
% of reference values [b]	77 (13)	76 (13)	81 (12)	
VO_{2peak} (mL/kg/min)	31.5 (6.3)	33.2 (6.2)	27.9 (4.8)	0.01 [a]
% of reference values [b]	88 (17)	87 (19)	90 (15)	
Peak power output (W)	144 (30)	153 (26)	124 (30)	0.003 [a]
Peak heart rate (BPM)	175 (17)	172 (117)	184 (13)	
% of predicted values (220-age)	96 (9)	94 (5)	100 (7)	
Peak RER	1.19 (0.12)	1.16 (0.11)	1.26 (0.12)	
Perceived exertion [c]	9.0 (1.7)	9.2 (1.8)	8.5 (1.3)	
Level of everyday PA				
Duration of dynamic activities (% of 24-hours)	8.6 (3.5)	8.4 (3.4)	9.1 (3.7)	0.54
% of reference values [d]	85 (35)	89 (36)	75 (31)	
Self-reported level of PA (MET hours/day)	14.8 (2.5)	14.4 (13.3)	15.8 (11.1)	0.75
Fatigue				
FSS score	4.1 (1.3)	4.1 (1.2)	4.0 (1.5)	0.74
Not fatigued (FSS score < 4.0)	50%	48%	54%	
Fatigued ($4.0 \leq$ FSS score < 5.1)	19%	21%	15%	
Severely fatigued (FSS score ≥ 5.1)	31%	31%	31%	

* p-values of independent samples t-tests for differences between men and women are pesented.

[a] Indicates a significant difference at $p \leq 0.05$.

[b] Reference values obtained from Dutch reference values (sedentary) [21].

[c] Perceived exertion as measured with Borg scale [22].

[d] Obtained from reference sample measured in our department.

BPM: beats per minute; RER: respiratory exchange ratio; PA: physical activity; MET: metabolic equivalent; FSS: Fatigue Severity Scale.

Level of everyday physical activity and fatigue

On average, objective PA was 8.6 (SD 3.5)%, which resembles 124 min of PA per day (Table 4.2). The participants were significantly less active than able-bodied age mates (10.9 [SD 3.5]%, $p < 0.01$). Self-reported PA was 14.8 (SD 2.5) MET hours/day. Thirty-one percent of all participants were severely fatigued (FSS score ≥ 5.1), and 19% of all participants experienced fatigue ($4.0 \leq$ FSS score < 5.1).

Relationships amongst physical fitness, level of everyday physical activity, and fatigue

Figure 4.1(a) shows the scatter plot for the relationship between physical fitness and objective PA; men are indicated by triangles and women by squares. There was no relationship between physical fitness and objective PA (men: $R_p = 0.26$, $p = 0.18$; women: $R_p = 0.26$, $p = 0.38$).

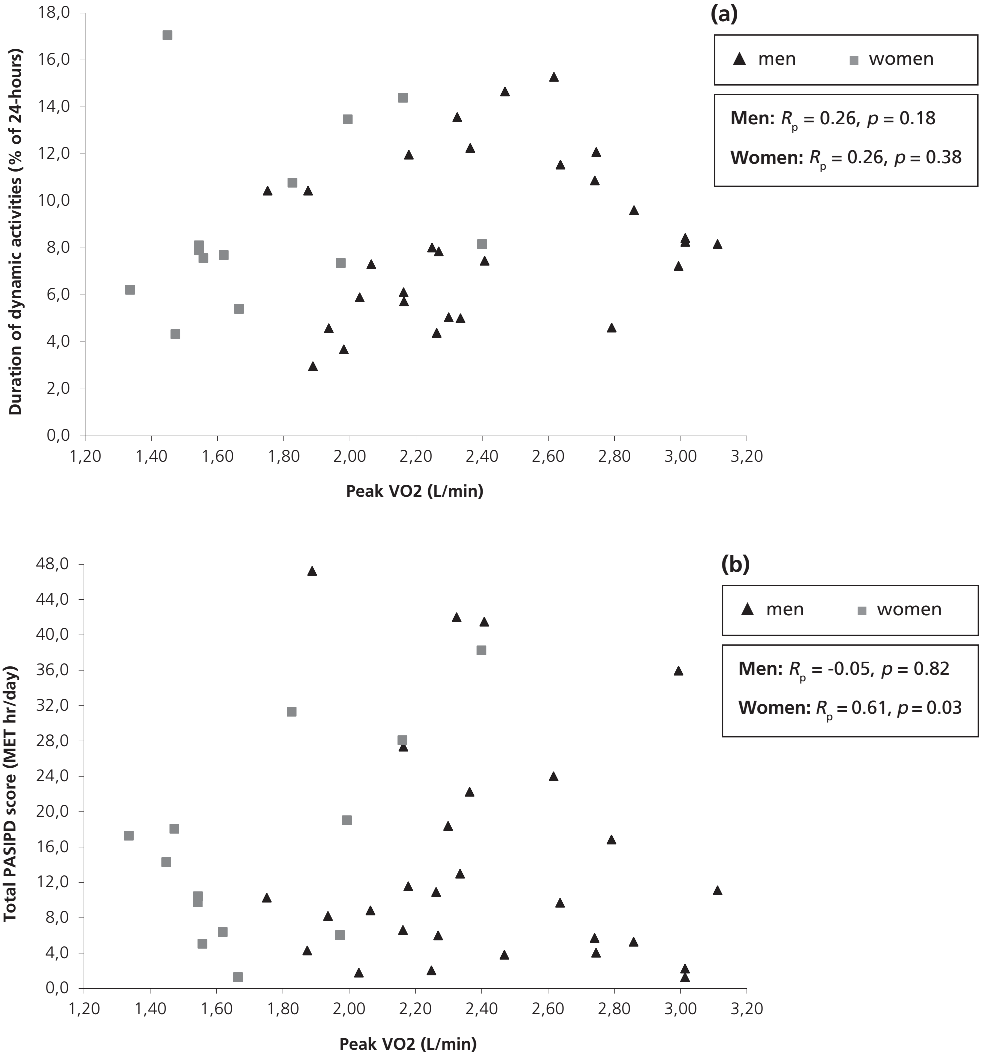

Figure 4.1 **(a)** Relationship between physical fitness and the objective level of physical activity. **(b)** Self-reported level of physical activity for men (▲) and women (■).
R_p: Pearson's correlation coefficient; PASIPD: Physical Activity Scale for Individuals with Physical Disabilities; MET: metabolic equivalent.

Women with CP who had higher levels of self-reported PA had higher levels of physical fitness ($R_p = 0.61$, $p = 0.03$) (Figure 4.1(b)).

With regard to fatigue, we found that men with higher levels of physical fitness reported less fatigue ($R_p = -0.37$, $p = 0.05$) (Figure 4.2). In women, no relationship was found. Relationships between objective PA and fatigue were not significant in both men ($R_p = -0.16$, $p = 0.42$) and women ($R_p = 0.29$, $p = 0.34$). Also, self-reported PA was not associated with fatigue ($R_p = -0.15$, $p = 0.44$ and $R_p = -0.13$, $p = 0.66$, respectively). Additional analyses with VO_{2peak} corrected for body mass (mL/kg/min) showed no significant relationships amongst physical fitness, objective PA, self-reported PA and fatigue.

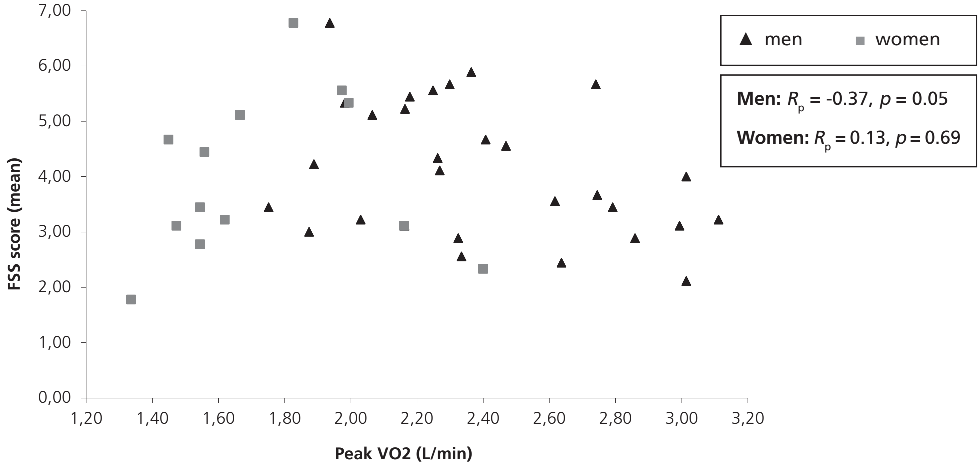

Figure 4.2 Relationship between physical fitness and fatigue for men (▲) and women (■). FSS: Fatigue Severity Scale; R_p: Pearson's correlation coefficient.

DISCUSSION

This is the first study to systematically describe the level of physical fitness and relationships with level of everyday PA and fatigue in a relatively large group of adults with CP. Participants had low levels of physical fitness, were less physically active than able-bodied age mates, and half of the participants experienced fatigue. In women, physical fitness was related to self-reported PA, and in men physical fitness was related to fatigue, indicating lower self-reported PA and higher fatigue associated with lower physical fitness levels.

Physical fitness

The low levels of physical fitness are in agreement with results found in earlier studies in adults [12,13] and in children and adolescents with CP [28-32] and could be attributed to a lower amount of active muscle mass, a reduction in localised muscle blood flow in the lower extremity muscles, or to deconditioning because of physical inactivity [14,28,33,34]. We did not find consistent relationships between physical fitness and the level of everyday PA; thus, we could not support a theory of deconditioning in which lower levels of PA lead to lower levels of physical fitness.

The low levels of physical fitness found in our population may improve with physical training, an aspect in which more (longitudinal) research is needed. The few studies on exercise programs have been shown to elicit favourable effects on physical fitness in both adults [35] and children with CP [30,32].

Relationships amongst physical fitness, level of everyday physical activity, and fatigue

We found a moderate positive relationship between physical fitness and self-reported PA in women. It appears that, in our sample, women who were more physically fit perceived themselves as being more physically active during the day. However, this was not confirmed in the objective measurement of PA or in men with CP. In other patient groups, results on relationships between physical fitness and the level of everyday PA are also inconclusive, with some studies reporting moderate relationships [36,37] and others reporting no relationships [38,39]. The lack of relations in our study between the level of everyday PA and physical fitness may be explained by a rather low intensity of everyday PA, and hence a low physical strain in the adults with CP, which may not have been high enough to induce changes in physical fitness. Future studies, in which this physical strain in daily life is determined with, for example, HR or oxygen uptake during everyday physical activities, could give us more insight in this possible mechanism.

Fatigue is important to consider in adults with CP, because it is highly prevalent in adults with bilateral CP and is suggested to be a cause of deterioration in, for example, walking function [9]. Severe fatigue was evident in 31% of participants in this study and 30% of participants in a Norwegian study [10]. We found that, although the relationship was not very strong, men with lower levels of physical fitness reported higher levels of fatigue. In women however, no relationship was found. Also, relationships between objective and self-reported PA and fatigue were not present, which is in line with other studies in adults with CP [10,40]. Opheim et al. [9] hypothesised that the fatigue experienced in their sample was of physical origin and could in part be attributed to low levels of physical fitness. However, we did not find conclusive evidence for a physical origin of the fatigue experienced in our sample. It appears that other factors may play a role in the existence of fatigue in CP. One could hypothesise that possibly the imbalance between physical fitness and the level of everyday PA may have a stronger influence on fatigue than physical fitness or PA itself. In addition, other factors such as behavioural factors should be included in a more psychological-oriented model of fatigue.

We need to take into account that this is a cross-sectional study and that we cannot infer causality in relationships amongst physical fitness, level of everyday PA, and fatigue. The apparent lack of consistent relationships amongst these three aspects might be a consequence of the relatively small sample size (29 men and 19 women), and other measures of physical functioning such as physical strain or energy expenditure may have yielded different results. However, the absence of strong relationships between physical fitness, PA, and fatigue is further amplified by the lack of significant relationships between VO_{2peak} corrected for body mass (mL/kg/min) and the other outcome parameters.

Limitations

Our results can be fully generalised only to ambulatory adults with bilateral CP, because one of the inclusion criteria was the ability to perform a cycle ergometer test. We might have overestimated the level of physical fitness and the level of everyday PA because of selection bias. Adults with CP interested in physical fitness (and therefore possibly having higher levels of fitness and PA) may have been more likely to participate than those with less interest and lower levels of physical fitness and PA. Furthermore, we compared values of aerobic capacity in adults with CP with reference values of Dutch sedentary persons. Those values were measured indirectly with submaximal tests [21], but other values were not present for the Dutch situation. Similar outcomes were found when we compared our results with reference values from Shvartz and Reibold [41], which were measured directly in untrained persons in the United States, Canada, and seven European countries. Finally, fatigue and self-reported PA were determined with (recall) questionnaires, which might have induced recall bias (self-reported PA) and socially desirable answers (fatigue and self-reported PA).

Perspective

Our study indicates that ambulatory adults with CP have low levels of physical fitness, are less physically active (both objectively measured and self-reported), and often experience fatigue. Theories of deconditioning are often suggested for people with physical disabilities in which impairments can lead to lower levels of PA, which may be related to lower levels of physical fitness and, in turn, to further diminished daily levels of PA. However, in our study, we did not find evidence for strong relationships amongst physical fitness, PA, and fatigue, and other mechanisms may play a role in explaining, for example, the existence of fatigue in CP. Nevertheless, interventions aiming to improve physical fitness and PA are warranted, and large-scale longitudinal studies may help us further determine which factors are responsible for the lowered overall health in adults with CP.

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5

Cardiovascular disease risk in adults with spastic bilateral cerebral palsy

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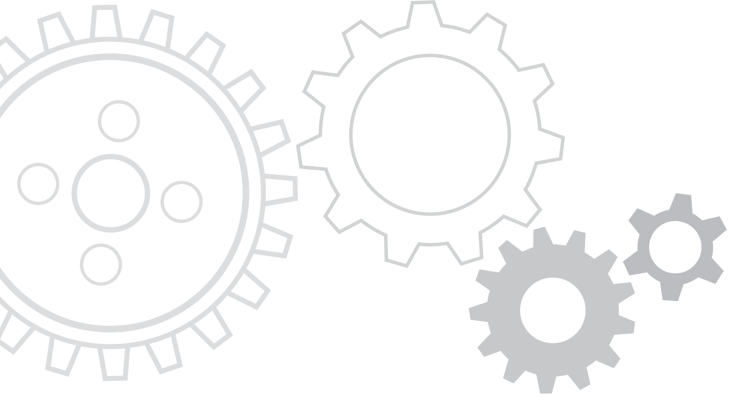

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ABSTRACT

Objective To explore 1) cardiovascular disease (CVD) risk factors and the 10-year clustered risk of a fatal cardiovascular event in adults with spastic bilateral cerebral palsy (SBCP) and 2) relationships between the 10-year risk and body fat, aerobic fitness and physical activity.

Design Cross-sectional study.

Participants Forty-three adults with SBCP without severe cognitive impairment (mean age 36.6 [SD 6] years; 27 men).

Methods Biological and lifestyle-related risk factors and the 10-year risk according to the Systematic Coronary Risk Evaluation (SCORE) were assessed. Relationships were studied using multivariable linear regression analysis.

Results The following single risk factors were present: hypertension ($n = 12$), elevated total cholesterol ($n = 3$), low HDL-C ($n = 5$; all men), high-risk waist circumference ($n = 8$), obesity (BMI; $n = 5$; all men), reduced aerobic fitness (on average 80% of reference values), reduced level of everyday physical activity (on average 78% of reference values) and smoking ($n = 9$). All participants had a 10-year risk ≤ 1%. Corrected for gender, participants with higher waist circumference ($\beta = 0.27$; $p = 0.06$) or BMI ($\beta = 0.25$; $p = 0.08$) tended to have a higher 10-year risk.

Conclusion In this relatively young adult sample of SBCP several single CVD risk factors were present. The 10-year fatal CVD risk was low and higher body fat tended to be related to higher 10-year risk.

INTRODUCTION

Cardiovascular disease (CVD) is a leading cause of morbidity and mortality worldwide, including in The Netherlands [1-3]. Numerous studies of the general population have demonstrated that high body fat, low aerobic fitness and lack of physical activity are risk factors for CVD [4-7]. Persons with chronic physical conditions such as cerebral palsy (CP), may be at increased risk of developing CVD [4] because they have low levels of both aerobic fitness [8,9] and everyday physical activity [10-12]. Nowadays, well-functioning persons with CP have a survival outlook close to that of the general population [13,14] and CVD may become of increasing concern. Strauss et al. [14] demonstrated that mortality due to ischemic heart disease and cerebrovascular disease were elevated in persons with CP, whereas Hemming et al. [13] found no such association.

Little is known about single CVD risk factors in CP. Some studies have been performed on body fat in adults, and conflicting results were found [15]. In the USA and Korea, the number of ambulatory children with CP and overweight has increased over time [16,17]. Reduced levels of physical fitness have been found amongst children [9] and adults [8,18] with CP. Low levels of everyday physical activity were demonstrated in children and adults with bilateral CP [11,12], but not in adults with unilateral CP [19]. Finally, Turk et al. [20] reported a low percentage of smoking (2%) amongst women with CP. To our knowledge comprehensive data concerning traditional CVD risk factors such as blood pressure, lipid profile and diabetes mellitus in adults with CP are scarce [21].

Exploring single CVD risk factors is necessary to obtain insights into condition-specific problems and preventive strategies. Apart from single risk factors, it is important to investigate clustered risk, because it estimates CVD risk better than the sum of separate risk factors [22]. In adults with CP, there are no objective data about clustered CVD risk.

The aims of the present study were to investigate biological and lifestyle-related CVD risk factors, and to assess the clustered 10-year risk of a fatal cardiovascular event in adults with spastic bilateral CP (SBCP), aged 25 to 45 years, without severe cognitive impairment. Furthermore, as body fat, aerobic fitness and everyday physical activity are modifiable factors from an exercise perspective, we explored associations between the 10-year risk and these factors. Because it is important to explore cardiovascular risk and begin preventive strategies early in life, a relatively young adult sample was studied [3,6].

METHODS

Study sample

Participants were recruited from ten rehabilitation centres in The Netherlands, including historical paediatric registers, and via the Association of Physically Disabled Persons and their Parents (BOSK). The inclusion criteria were a diagnosis of SBCP and age 25 to 45 years. Exclusion criteria were any multimorbidity with lasting effects on everyday physical activity or contraindicated for a progressive maximal ergometer test (e.g., severe cardiopulmonary disease), full dependence on electric wheelchair propulsion, inadequate Dutch language proficiency, legal inability and severe cognitive impairment according to medical files. The last criterion excluded persons who could not understand study instructions.

This study was part of a larger cross-sectional study on daily functioning and physical fitness. From the original cohort of 56 participants [23], 13 persons could not participate due to no consent to blood withdrawal ($n = 12$) and missing values for blood pressure ($n = 1$). Therefore, forty-three adults with SBCP participated in the present study. In a non-response study, participants were older than non-participants (mean difference 2.7 years; $p < 0.05$), but there was no difference regarding gender or distribution of limb impairment. All participants received verbal and written information about the study and gave their written consent to participate. The Medical Ethics Committee of the Erasmus Medical Center and the participating rehabilitation centres approved the study.

Measurements

Characteristics of study sample

Neuromotor abnormality was classified according to the Reference and Training Manual of the Surveillance of CP in Europe (SCPE) [24], and gross motor functioning according to the Gross Motor Function Classification System (GMFCS) [25]. Age, gender, ethnicity, level of education, employment and civil status were reported. For each participant the medical and family history of hypertension, CVD and diabetes mellitus, and the use of antihypertensive, lipoprotein metabolism and anti-diabetic drugs were recorded.

Biological risk factors

Blood pressure was measured while the participants were seated for 10 minutes prior to the measurement. Using a sphygmomanometer (Maxi Stabil, Speidel&Keller, Germany) and appropriately sized measure cuff, systolic blood pressure (SBP) and diastolic blood pressure (DBP) were measured twice and mean values were recorded. Blood pressure was classified according to the seventh report of the Joint National Committee on Prevention, Detection, Evaluation and Treatment of High Blood Pressure (JNC 7) [2].

Total serum cholesterol (TC), high-density lipoprotein cholesterol (HDL-C) and glucose were measured by taking nonfasting venous blood samples of approximately 10 ml from the vena antecubitis with a vacutainer. Their concentrations were determined using routine methods on a Roche Modular analyzer (Roche Diagnostics, Almere, The Netherlands). Values of lipids were categorised according to the National Cholesterol Education Programme, Adult Treatment Panel III (NCEP ATP III) [26] and glucose according to well known nonfasting cut points [3,27]. Nonfasting samples were used, which is allowed for CVD risk estimation [1,26,28].

As indicators of body fat we assessed waist circumference and body mass index (BMI). Waist circumference was measured midway between the lower rib margin and the iliac crest with the tape positioned evenly around the bare abdomen [1,27] while the participant was lying supine instead of standing. The mean waist circumference of two measurements was recorded in centimetres and classified according to Lean et al. [29]. BMI (kg/m^2) was calculated and classified according to the WHO criteria on BMI [1,3]. Height was measured while lying supine, in case of contractures from joint to joint. Weight was measured on a digital chair scale (Cormier, type H.F., France).

Aerobic fitness was measured during a progressive maximal exercise test on an electronically braked cycle ergometer (Jaeger ER 800, Jaeger Toennies, Breda, The Netherlands) by a portable breath-by-breath gas analyzer (K_4b^2, Cosmed, Rome, Italy). Detailed descriptions of this test, based on the McMaster All-Out Progressive Continuous Protocol, can be found elsewhere [8]. Aerobic fitness was operationalised as the mean oxygen uptake during the last 30 seconds of exercise (VO_{2peak}, expressed in L/min). The cycle ergometer test was performed in 35 participants. Eight participants were excluded because of severe hypertension ($n = 1$), missing values ($n = 2$) and inability to cycle ($n = 5$, due to severe spastic paresis, impaired balance and/or osteoarthritis).

Lifestyle-related risk factors

The level of everyday physical activity was objectively measured during two consecutive weekdays using an accelerometry-based activity monitor (AM; Temec Instruments BV, Kerkrade, The Netherlands) [11,30]. The level of everyday physical activity was operationalised as the average duration of dynamic activities (composite measure of the separately detected activities walking [including walking stairs and running], wheelchair propulsion, cycling, and general noncyclic movement]) of the two measurement days and was expressed in minutes per day.

Smoking behaviour and alcohol consumption were assessed in a face-to-face interview. We recorded smoking (yes/no), and classified the level as heavy smoking (≥ 20 cig/day) or not [27]. Drinking alcohol was classified as never, light (1 drink/day), intermediate (> 1 – < 3 drinks/ day) or heavy (≥ 3 drinks/day) [27].

European Systematic Coronary Risk Evaluation (SCORE)

Following the current European and Dutch guidelines for primary prevention of CVD, the individual 10-year absolute risk of a fatal atherosclerotic event including heart attack, stroke, or aortic aneurysm was estimated by using the SCORE [1,28,31]. The SCORE is a cardiovascular disease risk assessment model based on a large data set tested thoroughly on European data [22,28]. It takes an individual's overall risk profile into account and estimates the clustered 10-year CVD risk directly. The individual SCORE-risk was calculated for all participants according to the equation of the original reference [22], which includes the risk factors age, gender, TC, mean SBP, and cigarette smoking. The SCORE low-risk equation was used, which is applicable for regions with a low CVD risk in Europe, including The Netherlands [32].

Dutch reference samples

Blood pressure, TC, HDL-C, TC/HDL-C ratio, glucose, BMI, smoking and alcohol behaviour were compared with Dutch reference values obtained using comparable measurement procedures, e.g. nonfasting blood samples. From a total of n = 22,769 persons, different subgroups, all aged 20 to 49 years, were selected according to the risk factor studied [27]. Aerobic fitness was compared with reference values obtained from a healthy, untrained sample from the USA, Canada and seven European countries (including The Netherlands) [33] and expressed as a percentage of these reference values. Everyday physical activity was compared with reference values of healthy age-mates from our own database and measured with the same AM protocol (n = 45) [30], and expressed as a percentage of these reference values. Reference values are presented in Table 5.2 and compared with values of persons with SBCP in the discussion.

Statistical analysis

Data were analysed using SPSS, version 17.0 (SPSS Inc., Chicago, IL, USA). All categorised results were tested by χ^2 tests. Subgroup analyses were performed with the *t*-test for independent samples (gender) or with one-way analysis of variance for means and the χ^2 test for proportions (GMFCS level in three subgroups: I, II and III/IV). In case of skewed variables the Mann-Whitney *U* test or Kruskal-Wallis test were used. In Tables 5.2 and 5.3 the results are presented for gender but not for GMFCS level, because the latter showed significant differences for only two risk factors. *P*-values below 0.05 were considered statistically significant.

Linear regression analyses were used to study associations. SCORE, waist circumference, BMI, aerobic fitness and level of everyday physical activity were treated as continuous variables. Multivariable linear regression analyses were performed for exploring associations with SCORE low-risk (dependent variable). We conducted separate regression analyses for each of the following independent variables, one at a time and corrected for gender [1,22]: GMFCS level, waist circumference, BMI, aerobic fitness or everyday physical activity. To compare the influence of variables with a different scale, standardised regression coefficients (Beta, β) were calculated, expressing the influence of a shift of one SD of the variables on the outcome of interest. Beta, confidence intervals (95% CI) and explained variance (R^2; %) are reported.

RESULTS

Characteristics and medical history of the study sample

Characteristics of the study sample are presented in Table 5.1. None of the adults with SBCP had a medical history of CVD or diabetes mellitus. Two adults had a medical history of hypertension and both of them used antihypertensive drugs prior to the study. No participants used lipoprotein metabolism drugs. The family medical history was known for 39 participants, of whom 6 participants had positive kindred’s (6 CVD, 2 diabetes mellitus).

Table 5.1 Characteristics of the study sample with spastic bilateral cerebral palsy (n = 43)

Characteristics	*n*
Age in years, mean (SD)	36.6 (6)
Gender (men)	27
Ethnicity (Caucasian)	43
Limb distribution	
Diplegia	23
Quadriplegia	20
GMFCS, *n*	
Level I	11
Level II	22
Level III	8
Level IV	2
Level V	0
Level of education [a]	
High	15
Medium	18
Low	10
Student/employment status	
Student	2
Competitively employed	25
Sheltered employed	4
Disability pension, including volunteer work	12
Civil status	
Married/living together	9
Single	34

[a] Levels of education: Low: no education or elementary school and pre-vocational practical education; Medium: pre-vocational theoretical education and upper secondary vocational education (vocational high school); High: secondary education, higher education and university.
SD: standard deviation.

Biologic and lifestyle-related risk factors for CVD

Descriptive results of single biologic and lifestyle-related risk factors for the participants with SBCP and the Dutch reference samples are shown in Table 5.2. According to classifications of CVD risk factors [2,3,26] (Table 5.3), several risk factors were present in some of the participants: hypertension (n = 11); elevated levels of TC (n = 3); and high waist circumference (n = 8). Risk factors present only in men with SBCP were: low HDL-C (n = 5), representing a major risk; and obesity based on BMI (n = 5). All adults with SBCP had normal blood glucose levels. Levels of aerobic fitness (on average 79.7 [SD 12.8]% of reference values) and everyday physical activity (on average 78.4 [SD 35.9]% of reference values) were both reduced in the participants.

Subgroup analyses showed a significant difference between men and women with SBCP for waist circumference based on categories ($p = 0.02$) (Table 5.3). In addition to obesity, underweight was present only in men (Tables 5.2 and 5.3). Women with SBCP had significantly more favourable scores for DBP, HDL-C, TC/HDL-C ratio and VO_{2peak} (expressed as a percentage of reference values) than men with SBCP (Table 5.2). For GMFCS level, two differences were found: more study participants in GMFCS level II than in levels I and III/IV had hypertension ($p < 0.001$), and a lower level of everyday physical activity was noted in participants with GMFCS level III/IV (82.0 [SD 43.8] min/day) than with GMFCS level I (148.7 [SD 41.3 min/day]) ($p = 0.009$).

10-year risk and associations with body fat, aerobic fitness and everyday physical activity

All adults with SBCP had a SCORE risk of ≤ 1%, which equates to a low 10-year risk of fatal CVD (Table 5.2). The risk for women was lower than for men ($p = 0.001$). Corrected for gender, participants with a higher waist circumference tended to be more likely to have a higher 10-year risk ($\beta = 0.27$, $p = 0.06$) (Table 5.4). Using BMI in the multivariable analysis gave comparable results ($\beta = 0.25$, $p = 0.08$). No associations were demonstrated between VO_{2peak} or everyday physical activity and the 10-year risk.

DISCUSSION

This sample of adults with SBCP without severe cognitive impairment, aged 25 to 45 years, had a 10-year fatal cardiovascular risk of ≤ 1%. Although this is a low 10-year risk, several single biological and lifestyle-related risk factors were present in the relatively young study sample.

In total, 12 (28%) participants had hypertension, including one person with a medical history of hypertension, who used antihypertensive drugs prior to our study resulting in prehypertensive values. Prehypertensive values were found in another 44% of the participants, which makes them at increased risk for progression to hypertension [2]. The study sample had a higher blood pressure than a Dutch reference sample [27], including a twofold higher hypertension rate in men with SBCP. The high proportion of participants with (pre)

Table 5.2 Biologic and lifestyle-related risk factors of cardiovascular disease and 10-year fatal cardiovascular disease risk in adults with spastic bilateral cerebral palsy and Dutch reference samples

	Adults with CP ($n = 43$)		Men versus women with CP	Reference sample ($n =$ [a])	
	Men ($n = 27$)	Women ($n = 16$)	*p*-value	Men ($n =$ [a])	Women ($n =$ [a])
Biological risk factors					
Blood pressure					
Systolic (mmHg) (mean [SD])	126 (10)	126 (10)	0.98	122	113
Diastolic (mmHg) (mean [SD])	86 (11)	81 (6)	0.03	77	73
Hypertension Systolic ≥ 140 Diastolic ≥ 90 mmHg (*n* [%])	8 (30%)	3 (19%)	0.50	15%	8%
Blood lipid and lipoproteins (mean [SD])					
Total cholesterol (mmol/L)	4.7 (0.8)	5.0 (0.9)	0.18	5.0	4.9
High-density lipoprotein (mmol/L)	1.3 (0.4)	1.8 (0.5)	< 0.01	1.2	1.5
Total cholesterol/HDL ratio	3.8 (1.4)	2.9 (0.8)	0.005	4.6	3.5
Glucose (mmol/L) (mean [SD])	4.1 (0.8)	4.7 (1.1)	0.06	5.2	5.0
Body fat (mean [SD])					
Waist circumference (cm)	87.8 (16.2)	80.9 (9.9)	0.09	na	na
Body Mass Index (kg/m^2)	24.3 (6.0)	23.5 (3.0)	0.60	24.7	24
BMI ≥ 30 (%)	5 (18.5%)	0 (0%)	0.07	7.2%	8%
Aerobic fitness ($n = 35$) [b]					
VO_{2peak} (L/min) (mean [SD])	2.4 (0.4)	1.7 (0.3)	< 0.01	3.1 (0.14)	2.0 (0.14)
VO_{2peak} as percentage of reference values [c] (%)	76.1 (11.5)	86.5 (12.8)	0.02		

	Adults with CP ($n = 43$)		Men versus women with CP	Reference sample ($n =$ [a])	
	Men ($n = 27$)	Women ($n = 16$)	p-value	Men ($n =$ [a])	Women ($n =$ [a])
Lifestyle-related risk factors					
Physical activity (min/day) (mean [SD])	108.6 (49.3)	127.0 (59.4)	0.28	135.5 (46.1) [d]	175.0 (46.9) [d]
Physical activity as percentage of reference values (%)	81.9 (37.2)	72.6 (34.0)	0.42		
Current smoker (yes) (*n* [%])	7 (26%)	2 (13%)	0.30	39%	39%
Smoking behaviour (≥ 20 cig/day)	1 (4%)	0 (0%)	0.44	36%	32%
Alcohol drinking never (*n* [%])	11 (41%)	11 (69%)	0.08	9%	18%
Alcohol light drinking, 1 drink/day (*n* [%])	12 (44%)	4 (25%)	0.20	35%	57%
Alcohol intermediate drinking, >1 – <3 drinks/day (*n* [%])	4 (15%)	1 (6%)	0.4	37%	21%
Alcohol heavy drinking ≥ 3 drinks/day (*n* [%])	0 (0)	0 (0)	na	19%	4%
Clustered 10-year fatal CVD risk					
SCORE risk (%) (mean [SD])	0.19 (0.18)	0.06 (0.06)	0.001	na	na

[a] From a total of $n = 22{,}769$ persons of the Dutch general population, subgroups (aged 20–49 years) according to the risk factor studied were selected [27].

[b] Aerobic fitness was measured in 35 participants.

[c] According to Shvartz [33].

[d] Able-bodied age mates ($n = 45$, age ± 5 years) measured with the same AM protocol [11,30].

na: not available; SD: standard deviation; BMI: body mass index; CVD: cardiovascular disease risk; SCORE: Systematic Coronary Risk Evaluation.

Table 5.3 Categorised cardiovascular disease risk factors in adults with spastic bilateral cerebral palsy ($n = 43$)

	Men ($n = 27$)	Women ($n = 16$)	p-value
	n	n	
Blood pressure Systolic/Diastolic (mm Hg) [a]			
Normative < 120/80	7	5	0.31
Prehypertension 120–139/80–89	12	8	
Stage I hypertension 140–159/90–99	3	3	
Stage II hypertension ≥ 160/100	5	0	
Total cholesterol (mmol/L) [b]			
Desirable < 5.2	19	10	0.55
Borderline high 5.2–6.2	7	4	
High ≥ 6.2	1	2	
High-density lipoprotein (mmol/L) [b]			
High (no risk) ≥ 1.56	7	10	0.03
In between 1.04–1.55	15	6	
Low (major risk) < 1.04	5	0	
Glucose (mmol/L)			
Normal < 7.8	27	16	na
Impaired > 7.8 – < 11.0	0	0	
Indication Diabetes Mellitus >11.0	0	0	
Waist circumference (cm)			
Underweight: women: < 68, men: < 79	11	0	0.02
Normal: women: ≥ 68 – ≤ 80, men: ≥ 79 – ≤ 94	5	8	
Increased: women: > 80 – < 88 , men > 94 – < 102	7	4	
High risk: women: ≥ 88, men: ≥ 102	4	4	
Body Mass Index (kg/m^2)			
Underweight < 18.5	4	0	0.07
Normal ≥ 18.5 –24.9	13	13	
Overweight ≥ 25.0–29.9	5	3	
Obese ≥ 30.0	5	0	

[a] According to the seventh report of the Joint National Committee on Prevention, Detection, Evaluation and Treatment of High Blood Pressure [2].
[b] According to the ATP III definitions of the third report of the National Cholesterol Education Programme [26].
na: not available.

hypertensive values underlines the importance of regularly measuring and managing blood pressure from young adulthood onwards.

Obesity according to BMI was more often reported in men with SBCP than men of the reference group. A high waist circumference was present in 19% of the participants. The adults with SBCP had reduced levels of both aerobic fitness and everyday physical activity

Table 5.4 Multivariable analysis amongst adults with spastic bilateral cerebral palsy (n = 43): relationships between 10-year CVD risk (SCORE, dependent variable) and one independent variable (GMFCS, body fat, aerobic fitness or everyday physical activity) corrected for gender

		Multivariable analysis	
Independent variables n = 43	β	95% CI	R^2 (%)
Gender	- 0.42	- 0.70 – - 0.14	17.6
GMFCS	0.02	- 0.26 – 0.30	
Gender	- 0.36	- 0.63 – - 0.08	24.5
Waist circumference (cm)	0.27	- 0.01 – 0.55	
Gender	- 0.40	- 0.67 – - 0.13	23.8
BMI (kg/m^2)	0.25	- 0.02 – 0.52	
Gender	- 0.47	- 0.87 – - 0.07	21.1
VO_{2peak} (L/min) n = 35	- 0.01	- 0.40 – 0.37	
Gender	- 0.41	- 0.69 – - 0.13	17.8
Physical activity (min)	- 0.05	- 0.33 – 0.23	

β: standardised regression coefficient; CI: confidence interval; R^2: represents explained variance.

(expressed as a percentage of reference values). These findings suggest controlling of the single risk factors body fat, aerobic fitness and physical activity in individuals with SBCP.

In contrast to the aforementioned, all adults with SBCP had normal levels of glucose. Furthermore, in comparison with the Dutch reference sample [27], the lipid profile was more favourable in the study sample. Also less smoking and alcohol consumption were reported in adults with SBCP than the reference group. Alcohol use amongst the adults with SBCP was light or intermediate, not heavy. Therefore an increased risk of CVD based on alcohol consumption is unlikely.

Corrected for gender, participants with a higher waist circumference or BMI tended to have a higher 10-year fatal CVD risk. This trend is in line with results of other studies on body fat and CVD risk in the general population [1,6,7]. We found no evidence for associations between aerobic fitness or everyday physical activity and the 10-year risk. This absence of associations might be due to the relatively young age of the study sample and attendant low 10-year risk. Other possible explanations for the lack of associations are the relatively small sample size and the rather mildly affected sample (mainly GMFCS I and II, partly due to exclusion of persons dependent on electric wheelchair propulsion or unable to perform cycle ergometry). Inclusion of more severely affected persons might have produced broader ranges of aerobic fitness and/or everyday physical activity, as well as possible associations with the 10-year CVD risk. Finally, aerobic fitness and everyday physical activity could influence CVD risk through indirect effects not covered by SCORE, in view of the known interrelationships amongst CVD risk factors [2,5,34].

The reduced levels of aerobic fitness and everyday physical activity might play a role in the high proportion of participants with (pre)hypertensive values [2]. Considering this, we additionally explored modifiable factors associated with blood pressure by multivariable linear regression analyses. Mean DBP, but not mean SBP, was associated with waist circumference ($\beta = 0.34$, $p = 0.03$) and BMI ($\beta = 0.36$, $p = 0.02$) after correcting for gender. Again, no associations were demonstrated with aerobic fitness or everyday physical activity, in which methodological factors might play a role. The higher proportion of hypertensive values demonstrated in GMFCS level II is probably a coincidence, as this category contains the largest number of study participants.

In a meta-analysis of general populations, Williams et al. [35] reported that aerobic fitness and physical activity have significantly different relationships to CVD. The reductions in relative risk were nearly twice as great for aerobic fitness as for physical activity. In more recent studies of general populations, aerobic fitness [36] but not everyday physical activity [34] was predictive for a healthy CVD risk profile later in life. For instance, in a Dutch study by Twisk et al. [6], aerobic fitness during adolescence was related to a healthy CVD risk profile at the age of 32 years, whereas everyday physical activity was not. However, it should be noted that methodological aspects (e.g., using questionnaires to assess everyday physical activity) might have influenced the above findings.

Study limitations

The studied sample was relatively small. A larger sample would give a more precise estimate of CVD risk. However, we put maximal effort into attaining a representative sample, e.g. by extensive recruitment amongst a broad population. Gender, level of education, and GMFCS level were comparable to adults with SBCP without severe cognitive impairment in a representative cohort from the same geographic region [37].

Some caution is needed on the measurements. First, nonfasting blood samples were used for practical reasons, which may lead to an overestimation of the CVD risk. However, the levels of TC and HDL-C are minimally altered when measured in fasting or nonfasting blood [38]. Furthermore, there is growing evidence that HDL-C and TC/HDL-C ratio predict CVD when measured under nonfasting conditions [38,39]. For the associations of fasting or nonfasting TC with CVD conflicting results were found [38,39]. The Dutch guidelines on cardiovascular risk management recommend measurement of a fasting lipid profile and glucose, but allow non-fasting values for the estimation of CVD risk, including screening of the glucose level [1]. The SCORE steering committee of the European Society of Cardiology [28] and the NCEP guidelines [26] allow measurement of nonfasting TC and HDL-C as well. Thus, for the purpose of our study (estimation of CVD risk), non-fasting samples are allowed.

Second, our study included all risk factors according to Dutch guidelines [1], except dietary pattern, which somewhat hampers interpretation of our findings. Third, height and waist circumference were measured in all participants while lying supine so as to obtain accurate measurements in case of deformities of the limbs or restrictions in standing. This may have resulted in an underscoring of abdominal obesity and height and may partly explain the higher proportion of men who were underweight based on waist circumference than on BMI.

Recently, Peterson et al. [21] demonstrated that waist-to-hip ratio was associated with several markers for dyslipidemia in adults with CP whereas BMI and waist circumference were not associated with these markers. Furthermore, it was suggested that BMI may be healthy in persons with CP who have excessive body fat due to muscle atrophy and diminished bone density and requires other cut points. Waist circumference was classified according to Lean et al. [29] because no classification exists for measurements recorded in the supine position. Considering the problems of measuring waist circumference and BMI in CP, both were used in the multivariable analyses and showed comparable results.

Finally, other parameters (e.g., energy expenditure, sedentary time) or longer durations of measurement might deepen our understanding of everyday physical activity and its potential association with CVD risk. However, our aim was not to characterise habitual physical activity but to obtain insight into the level of everyday physical activity. Therefore, monitoring during two randomly selected weekdays in both persons with CP and the reference sample is considered to be an adequate duration to reliably record activities [30,40].

Strengths of the study are the extensive objective measurements of CVD risk, unlike in other studies where questionnaires are often used, e.g. to assess physical activity. The study fills a gap in the literature on biological and lifestyle-related CVD risk factors and the clustered CVD risk in adults with CP.

In conclusion, in this sample of relatively young adults with SBCP the 10-year fatal CVD risk was low, but several single CVD risk factors were present, of which (pre)hypertensive blood pressure was prominent. A higher level of body fat tended to be related to a higher 10-year risk, but no relations were demonstrated with aerobic fitness or everyday physical activity.

Further research is warranted with other measures (e.g., sedentary time, energy expenditure) and/or a different sample (more severely affected persons, older ages) to get more insight in CVD risk and its relationships with modifiable factors. Thus far, the current findings highlight the importance of screening for CVD risk factors in SBCP, specifically blood pressure and body fatness, and to start preventive strategies from young adulthood onwards.

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6

Participation and health-related quality of life in adults with spastic bilateral cerebral palsy and the role of self-efficacy

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ABSTRACT

Objective To assess participation and health-related quality of life (HRQoL) in adults with spastic bilateral cerebral palsy (CP), and to explore associations with self-efficacy.

Design Cross-sectional study.

Participants A sample of 56 adults with spastic bilateral CP (mean age 36.4 [SD 5.8] years; 62% men).

Methods Daily activities and social participation (LIFE-H 3.0), HRQoL (SF-36 Health Survey), demographic and clinical characteristics, and self-efficacy (General Self-Efficacy Scale [GSES-12]) were assessed. Associations were studied using multivariable logistic regression analyses.

Results At least 60% of the sample had difficulties with mobility, recreation and housing, and 44% had difficulty with personal care and employment. They perceived a low HRQoL for physical functions, but not for mental functions. Corrected for demographic and clinical characteristics, general self-efficacy explained 49% of the variance in outcome on social participation, and the subscale Effort (GSES-12) explained 32% of the variance for the physical HRQoL and 16% for the mental HRQoL.

Conclusion A significant number of adults with spastic bilateral CP encountered difficulties in social participation and had a low perceived HRQoL for physical functions. Higher general self-efficacy or a greater willingness to expend effort in achieving a behaviour was related to better participation and a higher physical and mental HRQoL.

INTRODUCTION

In the past few years, adults with cerebral palsy (CP) have gained increasing attention [1-4]. In general, emphasis has been placed on health issues and functional deterioration. Poor health may result in restrictions in activities and participation [2,5,6] and may affect quality of life [6-8]. Participation and quality of life can be considered as relevant outcomes with regards to functioning. This study examines these outcomes in adults with CP and investigates associated factors.

According to the International Classification of Functioning, Disability and Health (ICF), participation is defined as the involvement of a person in life-situations and is determined by impairment or limitations of activity, as well as by environmental or contextual factors [9,10]. For adults with CP, restrictions in participation have been demonstrated in several areas of day-to-day living, including employment, independent living, and social and leisure activities [1,5]. Adults with unilateral CP performed relatively well in these areas [11], indicating less restricted participation than adults with bilateral CP [1]. Objective information about adults with bilateral CP is scarce. The present study focuses on adults with bilateral CP, who seem to be more at risk of restrictions to participation.

Quality of life is defined as the individual's perception of their position in life in the context of the culture and value systems in which they live, and in relation to their goals, expectations, standards and concerns [12]. Health-related quality of life (HRQoL) refers to those aspects of life that are related to a person's health status. Little is known about the standard of HRQoL in adults with CP. Two studies have been published so far; Gaskin and Morris [13] studied HRQoL in relation to physical activity in an Australian CP population and demonstrated that, in general, the patients had low levels of physical functioning and vitality. Jahnsen et al. [8] reported that their results in most domains of HRQoL were significantly lower in adults with CP than those in a Norwegian reference sample.

For both outcomes of functioning, associated factors were assessed in previous studies, and a few of these also explored associations with self-efficacy. The concept of self-efficacy was introduced by Bandura [14] and is considered to be the belief of a person in his or her ability to organise and execute behaviours that are necessary for producing outcomes that the person desires. Self-efficacy does not reflect a person's skills, but rather a person's judgement of what they can do with their skills. There is evidence that self-efficacy is an important predictor for academic success, motivation and the functional well-being of people with disabilities, and that it is improvable by psychosocial interventions such as mentoring [15]. In young adults with a disability, including CP, a higher level of self-efficacy was associated with participation [16]. In the literature, several other associated factors for participation were addressed, including demographic factors, CP-related characteristics and environmental factors. Severe cognitive impairment, a low level of education, severe physical disability, and epilepsy were regarded as negative factors for being competitively employed and living with a partner [2,6,17]. For HRQoL, physical functioning [13], pain and fatigue [4,7] seemed to be important associated factors in adults with CP. Knowledge of associated factors is a prerequisite for the development of interventions needed to successfully rehabilitate patients with CP.

The aims of the present study were to study participation and HRQoL in adults with spastic bilateral CP without severe cognitive impairment, and to explore the factors associated with social participation and HRQoL, focusing in particular on self-efficacy.

METHODS

Study sample

Participants were recruited from ten rehabilitation centres in the western and central regions of The Netherlands and, via the Association of Physically Disabled Persons and their Parents (BOSK). Historical registers of paediatric rehabilitation and current registers of adult rehabilitation were used to trace persons with CP. A letter with information on the study and invitation to participate were sent to the persons with CP. To optimise the response a reminder was sent 4 weeks later.

The inclusion criteria were: spastic bilateral CP in a person aged between 25 and 45 years at the time of inclusion. Exclusion criteria were: comorbidities with lasting effects on physical activity, contraindications for a progressive maximal cycle ergometer test (e.g., severe heart disease), full dependence on electric wheelchair propulsion, legal inability to participate, inadequate comprehension of the Dutch language and cognitive impairment preventing understanding the study instruction. This study is part of a larger one, and some of the inclusion criteria were required for measuring physical activity and fitness, which were part of that study [18].

Of a total of 208 eligible persons with CP, 56 participated (response rate 27%). Twenty-four percent did not respond to the initial invitation to join the study, and 10% could not be reached due to a change of address. Another 39% refused to participate, of which 61% gave a reason for non-participation: burden of the research, problems with the duration of assessment, a lack of time (54%); personal reasons that discourage them from participating, comorbidities and communication problems (28%); and no interest (18%). On average, participants were older than non-participants (mean difference 2.5 years; *t*-test, $p < 0.01$), but did not differ regarding gender or the distribution of impaired limbs [18].

All participants received verbal and written information about the study and gave their written consent to participate. This study was approved by the Medical Ethics Committee of the Erasmus MC and all of the participating rehabilitation centres.

Measurements

Data were collected in face-to-face interviews and/or with standard questionnaires administered in the presence of a trained researcher and by physical examination or functional tests. Because of face-to-face measurements there were only a few missing values (see Tables).

The following characteristics were determined: age, gender, level of education, neuromotor abnormality, gross motor functioning and functional level (see Table 6.1). Level of education was used as an indicator of cognitive functioning. We distinguished three levels of education: 1) low, including no education, elementary school and pre-vocational practical education; 2) medium, including pre-vocational theoretical education and upper secondary vocational education; 3) high, including secondary non-vocational education, higher education and university. A physical examination was performed to assess the neuromotor abnormality. The classification system described in the Reference and Training Manual of the Surveillance of CP in Europe (SCPE) was used to classify persons with CP [19]. Gross motor functioning was classified according to the Gross Motor Function Classification System (GMFCS), which is a five-level classification system graded by the age-related severity of gross motor limitations [20]. Distinctions between levels of motor functioning are based on functional limitations and the need for assistive devices [20]. The GMFCS was originally developed and validated for children, but it has been shown to be reliable and valid for describing gross motor functioning in adults with CP [21].

The functional level was used to describe the study sample. This was assessed with a semi-structured interview based on the Functional Independence Measure (FIM), which includes 18 items that assess functioning in basic physical and cognitive abilities. Each item is scored on a scale of seven-levels from 1 (total assistance required) to 7 (complete independence). The items are grouped into a motor dimension (maximum score 91), a cognitive dimension (maximum score 35) and a total score (maximum score 126) [22]. The FIM has been used in other studies of CP, and has excellent inter-rater and test-retest reliability intercorrelation coefficient (ICC 0.95) [22,23].

Activities and participation

The short version of the Assessment of Life Habits (LIFE-H 3.0) was used to evaluate functioning in daily activities and social participation. The LIFE-H 3.0 assesses performance in the accomplishment of daily activities and social roles, and the overall satisfaction with performance [24]. The LIFE-H is composed of 69 life habits divided into 12 categories. The categories nutrition, fitness, personal care, communication, housing and mobility refer to daily activities; the categories responsibilities, interpersonal relationships, community life, education, employment and recreation address social roles. The performance score is based on two concepts: the level of difficulty when performing a life habit, and the type of assistance required to perform the habit. Both elements are combined in a scale ranging from 'not accomplished or achieved' (0), indicating total handicap to 'accomplished without difficulty' (9), indicating optimal activity or participation. Specific life habits that were not part of a respondent's lifestyle because of personal choice were marked as 'not applicable'. Mean scores were calculated for each category, for the daily activities and social participation sub-domains and total score. We regarded a mean score < 8.0 on the LIFE-H as representing difficulties [25].

The LIFE-H comprises a second scale that evaluates the individual's level of satisfaction with the accomplishment of life habits. Satisfaction is rated on a five-point Likert scale; varying from very dissatisfied (1) to very satisfied (5).

The LIFE-H has been shown to be reliable for different groups of adult patients, also the Dutch language version of LIFE-H [26]. The reliability and validity of the satisfaction scale have not yet been studied.

In addition to the LIFE-H, we assessed several indicators of participation. These were related to employment status, civil status, living status and whether or not the respondents had children (see Table 6.2).

Health-related quality of life

Health-related quality of life was assessed using the Short Form-36 Health Survey (SF-36). The SF-36 is designed to measure eight health domains on the basis of the participant's perceived burden of his or her illness: physical functioning, role limitations due to physical health problems, bodily pain, general health, vitality, social functioning, role limitations due to emotional problems and mental health. A composite score for the SF-36 domains was calculated from the raw scale scores using a z-score transformation (score range of 0–100) [27]. In addition, scores were summarised by two scale scores: a physical component summary scale (PCS) and a mental component summary scale (MCS), which were expected to have a mean of 50 (SD 10) in the general population of the USA [27]. These scales are used as a measure of participants' overall HRQoL. Higher scores indicate a better HRQoL. Psychometric evaluation and reference values were available for the Dutch population [28,29].

Self-efficacy

We assessed the participants' self-efficacy by using the Dutch language version of the General Self-Efficacy Scale-12 (GSES-12) [30,31]. The scale consists of 12 items that are scored on a five-point Likert scale. The scale is divided in three subscales: 1) Initiative, willingness to initiate behaviour; 2) Effort, willingness to expend effort in completing the behaviour; and 3) Persistence, defined as persistence when facing adversity. The total scale and subscales showed satisfactory levels of internal consistency (Cronbachs' alpha 0.63–0.69) [30] and test-retest reliability ($R = 0.67$) [32].

Statistical analysis

Analyses were performed using SPSS 12.0.1 for Windows. Results were described by descriptive statistics.

Indicators of participation were compared with the Dutch population, by using data available from the Statistics Netherlands (for years 2005–2007) [33], with a binomial test.

One sample *t*-tests were used 1) to compare scores of the SF-36 with Dutch reference groups: a group aged 16–40 years for domain scores [28], and a group aged 20–49 years for PCS and MCS scores [29]; and 2) to compare scores of the GSES-12 with a Dutch reference group, mean (SD) age 46.9 (18.1) years [34]. For all analyses, p-values ≤ 0.05 were considered to be significant.

To study associated factors of participation and HRQoL, univariable and multivariable logistic regression analyses were performed. Univariable analyses addressed relationships between demographic and clinical characteristics (age, gender, gross motor functioning, level of education and general self-efficacy) and indicators of participation, participation and HRQoL. For four dependent variables we explored multivariable models: 'daily activities' and 'social participation' of the LIFE-H and physical and mental HRQoL (PCS and MCS). Accommodating to the number of independent variables we included level of education, gross motor functioning and self-efficacy (General scale and Effort subscale) as independent variables. Because the dependent variables were not normally distributed the scores were dichotomised. For the sub-domains 'daily activities' and 'social participation' of the LIFE-H, individuals were classified as either having difficulties (0) or no difficulties (1). For the PCS and MCS of the SF-36, a mean score < 50.0 indicated a lower level of HRQoL (0) and a mean score ≥ 50.0 demonstrated a higher level of HRQoL (1). Of the independent variables, the GMFCS level was categorised in three groups: I, II, III/IV; none of the participants had a GMFCS level V. Because clinically relevant cut-off points for good and poor GSES-12 total and subscale scores are unknown, we dichotomised these scores based on the median of the study sample, identifying persons with relatively low (0) and high scores (1) [35]. Odds ratios

Table 6.1 Characteristics of the study sample (*n* = 56)

Characteristics	
Age in years, mean (SD)	36.4 (5.8)
Gender, *n* (%)	
Men	35 (62)
Women	21 (38)
GMFCS, *n* (%)	
Level I	13 (23)
Level II	28 (50)
Level III	11 (20)
Level IV	4 (7)
Level V	0 (0)
Level of education, *n* (%)	
High	17 (30)
Medium	24 (43)
Low	15 (27)
Functional level based on the FIM, mean (SD)	
Subscale scores (maximum score)	
Motor dimension (91)	83.9 (8.7)
Self-care (42)	38.5 (5.5)
Sphincter control (14)	13.9 (0.3)
Transfers (21)	19.4 (2.2)
Locomotion (14) [a]	12.2 (1.7)
Cognitive dimension (35)	33.1 (2.0)
Communication (14)	13.2 (1.0)
Social cognition (21)	19.9 (1.3)
Total score (126)	117 (9.4)

[a] 1 woman missed the question on mobility.
FIM: Functional Independence Measure; SD: standard deviation; GMFCS: Gross Motor Function Classification System.

(OR), including 95% confidence intervals (95% CI), and explained variances (R^2, Nagelkerke R square) of the regression models are presented.

RESULTS

Characteristics of the study sample

Table 6.1 presents the personal characteristics and functional level of the 56 adults with bilateral CP who participated in this study. The mean age was 36.4 (SD 5.8) years and 62% were men. Seventy-three percent of the participants had a high level of gross motor functioning (GMFCS level I or II). Seventy-three percent had a medium or high level of education. Participants had a high degree of independence in performing functional activities according to the FIM, with a mean total score of 117 (SD 9.4) out of a maximum of 126.

Daily activities and participation

The indicators of participation as shown in Table 6.2. Sixty-eight percent of the adults with CP were in paid employment, including 54% who were in competitive employment and 14%

Table 6.2 Indicators of participation

	n (%)
Student/employment status	
Student	2 (4)
Competitively employment	30 (54)
Sheltered job	8 (14)
Volunteer job	6 (11)
Unemployed	1 (2)
Household	0 (0)
Disability pension	9 (16)
Civil status	
Living together/married	15 (27)
Single	41 (73)
Living status	
Living with partner/others	16 (28)
Living alone	34 (61)
Living with parents	4 (7)
Living in an institution	2 (4)
Having children	6 (11)

in sheltered employment. In comparison, 77% of the overall Dutch population, aged 25–45 years, had paid employment ($p = 0.08$) [33]. Of the adults with CP, 27% were married or living with a partner. This percentage is low compared with a cohort of age-matched Dutch people (70%) ($p < 0.001$). A difference was also found with regards to having children: 11% of the adults with CP had children compared with approximately 50% of Dutch households in the 25–45 years age group ($p < 0.001$) [33].

The mean scores of daily activities and social participation as measured with the LIFE-H are shown in Table 6.3. According to the high mean scores in most categories, the participants function relatively well. However, according to the variance around the mean scores, a substantial number of subjects are restricted in their activities and social participation. In Figure 6.1, the percentage of participants with difficulty in each LIFE-H category is presented. With regards to daily activities, most adults with CP encountered difficulties in mobility (77% of the participants) and housing (71%) despite the use of assistive devices, adaptations or human assistance. Assistive devices used for mobility were a wheelchair (54%), a tricycle (39%), a scoot mobile (21%), a moped (7%) or a handbike (2%). Approximately half of the participants used a car, of which 11% was adapted.

Regarding housing, 36% of the participants had a house with adaptations, 7% lived in a house with outside assistance on call, and 4% lived in an institution. Informal caregiver assistance

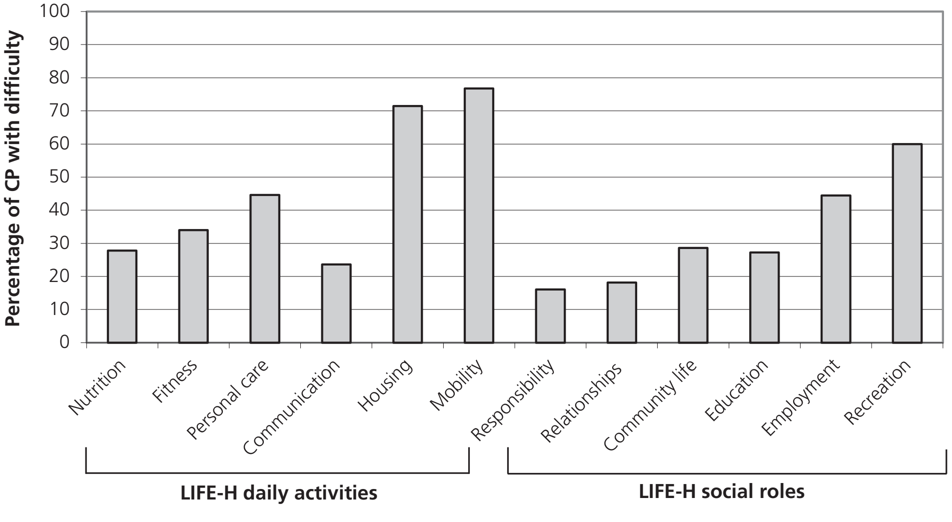

Figure 6.1 Percentage of adults with cerebral palsy (CP) with difficulty in each Assessments of Life Habits (LIFE-H) category.

Table 6.3 Functioning in activities, participation and health related quality of life

	Mean (SD)	Mean (SD)	
Functioning (LIFE-H)	Performance (0–9)	Satisfaction (1–5)	
Daily activities			
Nutrition	8.0 (1.5)	4.2 (0.5)	
Fitness	8.0 (1.2)	4.1 (0.6)	
Personal care	7.7 (1.5)	4.1 (0.5)	
Communication	8.3 (1.0)	4.1 (0.5)	
Housing	6.9 (1.2)	4.1 (0.5)	
Mobility	6.8 (1.4)	3.9 (0.6)	
Daily activities sub-domain	7.5 (1.0)		
Social participation			
Responsibility	8.6 (0.9)	4.2 (0.5)	
Relationships	8.5 (0.9)	3.4 (0.5)	
Community life	8.0 (1.4)	4.0 (0.5)	
Education [a]	8.4 (0.9)	3.9 (0.7)	
Employment [a]	7.8 (1.3)	4.1 (0.6)	
Recreation	7.2 (1.6)	4.1 (0.6)	
Social roles sub-domain	8.0 (1.0)		
Total score	7.7 (0.9)		
HRQL (SF-36) (0–100)	CP ($n = 56$)	Reference values [b]	p-value [d]
Physical functioning	60.1 (24.7)	93.1 (11.8)	< 0.001
Physical role function	68.8 (37.3)	86.4 (27.6)	0.001
Bodily pain [c]	74.3 (23.4)	80.9 (19.4)	0.04
General health	72.7 (17.3)	78.2 (17.3)	0.02
Vitality	62.5 (18.7)	70.7 (16.4)	0.002
Social funcioning	81.7 (20.4)	87.8 (19.1)	0.03
Emotional role function	81.5 (30.4)	85.4 (30.0)	0.35
Mental health	74.1 (18.6)	78.7 (15.2)	0.07
	CP	Reference values [e]	p-value [d]
Physical Component			
Summary Men [c]	45.8 (8.7)	52.5	< 0.001
Summary Women	41.4 (8.6)	51.6	< 0.001
Mental Component			
Summary Men [c]	52.1 (10.3)	49.1	0.10
Summary Women	53.7 (11.5)	46.7	0.01

[a] The category education was applicable for 11 participants and employment for 27.

[b] Dutch population aged 16 years and over ($n = 1742$), subgroup 16–40 years [28].

[c] Two missing values for bodily pain (men).

[d] p-value of one-sample t-test.

[e] Dutch population aged 20–49 years ($n = 22{,}769$) [29].

CP: cerebral palsy; LIFE-H: Assessment of Life Habits; SD: standard deviation; HRQoL: health-related quality of life; SF-36: Short Form-36 Health Survey.

was used by 16% and professional household assistance by 32%. With regards to personal care, 45% of the adults with CP had difficulties and at least 4% used professional assistance.

Regarding social participation, 60% of the participants had difficulty in recreational activities and 44% with employment. Difficulty in daily activities and social participation did not differ between men and women with CP. Only the category 'mobility' showed a significantly lower mean score for women with CP than men, 6.2 (SD 1.6) versus 7.1 (SD 1.1) ($p = 0.015$).

Participants were mostly satisfied with their performance of daily activities and social participation on the LIFE-H (Table 6.3). They were the most dissatisfied with their interpersonal relationships, with a mean score of 3.4 (0.5) of a maximum of 5.

Health-related quality of life

The HRQoL of the study sample is shown in Table 6.3 and compared with Dutch reference values [28,29]. Adults with CP perceived a significantly lower HRQoL for most domains, except for role limitations due to emotional problems and mental health. Regarding the physical component summary scale (PCS), participants perceived a relatively low HRQoL. On the mental component summary scale (MCS) women with CP had a better score (53.7 [SD 11.5]) than women in the reference group (46.7 [$p = 0.01$]). No difference was found between men in the two groups. Between men and women with CP, no significant differences were found.

Self-efficacy and its associations with daily activities, participation and health-related quality of life

General self-efficacy did not differ between adults with CP and a Dutch reference group, with a total score of 47.0 (SD 6.3) vs 46.3 (SD 7.7) ($p = 0.44$) (see Table 6.4). Adults with CP scored significantly better on the persistence subscale than did the reference group (21.3 [SD 2.8] vs 19.8 [SD 3.3], [$p < 0.001$]).

For the indicators of participation presented in Table 6.2 no correlations were found by univariable regression analyses with age, gender, gross motor functioning, level of education and general self-efficacy, except for employment, which was associated with age (OR = 0.8; $p = 0.001$, $R^2 = 30\%$). However, in multivariable models these determinants were not significant. In multivariable analyses, a higher level of gross motor functioning appeared to be positively associated with both daily activities (OR = 4.6; $p \leq 0.01$) and social participation (OR = 10.5;

Table 6.4 Self-efficacy

Self-efficacy (GSES-12, maximum score)	CP ($n = 56$)	Reference values ($n = 330$) [a]	p-value [b]
Effort (20)	14.7 (3.3)	15.5 (3.8)	0.07
Persistence (25)	21.3 (2.8)	19.8 (3.3)	< 0.001
Initiative (15)	11.0 (3.0)	11.0 (3.1)	0.93
Total (60)	47.0 (6.3)	46.3 (7.7)	0.44

[a] Dutch population aged 46.9 (18.1) years, $n = 330$ [32].
[b] p-value of one-sample t-test.
GSES-12: General Self-Efficacy Scale-12; CP: cerebral palsy.

$p \leq 0.01$). In addition, general self-efficacy contributed to social participation (OR = 6.2; $p \leq 0.05$). The multivariable models explained 40% of the total variance of functioning in daily activities and 49% in social participation (Table 6.5).

For the physical and mental HRQoLs, we found that corrected for demographic and clinical characteristics the self-efficacy Effort subscale explained 32% and 16% of the variance in outcome, respectively (Table 6.6). Participants with better gross motor functioning were 6 times more likely to perceive a higher physical HRQoL ($p \leq 0.01$). Participants with a higher score on the Effort subscale of GSES-12 were 7.6 times more likely to perceive a higher physical HRQoL ($p \leq 0.05$) and 3.5 times more likely to perceive a higher mental HRQoL ($p \leq 0.05$). Age, gender, level of education and general self-efficacy were not associated with mental and physical HRQoL.

DISCUSSION

Participation, health-related quality of life and associated factors

The present study investigated participation and HRQoL in adults with spastic bilateral CP without severe cognitive impairment. We found that, regarding employment, civil status, and having children, adults with CP are in a disadvantaged position compared with the Dutch reference population. Being involved in participation, varies between this study and that reported in other recent studies carried out in Scandinavian countries and the USA, which also focused on adults with CP without severe learning disabilities [1,7,17]. Although these study samples differed regarding neuromotor abnormality and distribution of limb impairment, they provide the best available reference descriptions of participation in adults with CP. For

Table 6.5 Basic characteristics and self-efficacy associated to daily activities and social participation (LIFE-H)

	Daily activities						Social participation					
	Univariable			Multivariable [a]			Univariable			Multivariable [a]		
	OR	95% CI	R^2	OR	95% CI	R^2	OR	95% CI	R^2	OR	95% CI	R^2
Basic characteristics												
GMFCS (I, II, III–IV)	4.9***	1.8–13.5	28%	4.6***	1.5–14.2		7.5***	2.3–23.9	36%	10.5***	2.5–44	
Level of education (low, middle, high)	3.5***	1.5–8.3	22%	2.4*	0.9–6.2		2.5**	1.1–5.5	13%	1.5	0.6–3.9	
Self-efficacy												
GSES-12 (total score)	2.4	0.8–7.3	6%	3	0.8–11.9	40%	3.1**	1–9.9	10%	6.2**	1.3–30	49%

*** $p \leq 0.01$
** $p \leq 0.05$
* $p < 0.10$
[a] Enter model.
LIFE-H: Assessment of Life Habits; OR: odds ratio; CI: confidence interval; R^2: Nagelkerke *R* square, represents explained variance; GMFCS: Gross Motor Function Classification System; GSES-12: General Self-Efficacy Scale-12.

Table 6.6 Basic characteristics and self-efficacy associated to Health Related Quality of Life (SF-36)

	Physical component summary scale						Mental component summary scale					
	Univariable			Multivariable [a]			Univariable			Multivariable [1]		
	OR	95% CI	R^2	OR	95% CI	R^2	OR	95% CI	R^2	OR	95% CI	R^2
Basic characteristics												
GMFCS (I, II, III–IV)	3.3**	1.2–8.7	17%	6.1***	1.7–21.7		1.0	0.5–2.3	0%	0.9	0.3–2.5	
Level of education (low, middle, high)	1.2	0.6–2.7	1%	0.5	0.2–1.5		1.9	0.8–4.2	6%	1.7	0.7–4.4	
Self-efficacy												
GSES-12 (Effort)	3.4*	0.8–14.1	9%	7.6**	1.4–42.4	32%	4.1**	1.2–14.0	13%	3.5**	1.0–12.8	16%

*** $p \leq 0.01$
** $p \leq 0.05$
* $p < 0.10$
[a] Enter model.
SF-36: Short Form-36 Health Survey; OR: odds ratio; CI: confidence interval; R^2: Nagelkerke R square, represents explained variance; GMFCS: Gross Motor Function Classification System; GSES-12: General Self-Efficacy Scale-12.

example, 54% of the adults in our study were found to be competitively employed compared with 33% in the study by Jahnsen et al. [7]. In addition, 14% of our subjects were engaged in sheltered employment compared with 18% in the study by Murphy et al. [17]. Previous studies have shown that adults with unilateral CP are more often competitively employed than those with bilateral CP [1,6,11]. Sixteen percent of subjects in the present study reported receiving a disability pension compared with 46% in Jahnsen et al. [7]. Difference in age between the study group of Jahnsen [7] and our sample might be of relevance in these figures on employment and disability pension, which is underlined by the association between age and employment in our sample.

The percentage of adults with CP who are married or living with a partner varies from 14% [1] to 27% (this study). Several aspects might possibly contribute to this low percentage of being married or living with a partner: a negative perception of the body [36], limited participation together with able-bodied persons (e.g., special education, sheltered employment), and more restricted to activities at home. Wiegerink et al., showed that Dutch young adults with CP had less experience with dating, romantic relationships and sexuality compared with Dutch age mates [37]. They also demonstrated the importance of participating in peer group activities for having a partner or sexual relationships [38]. Rates of living independently vary from 65% [7] to 90% (this study), and rates of living with parents range from 7% (this study) to approximately 33% [17]. The percentage of participants who have children ranges from 4% [17] to 11% (this study), with an outlying exception of 22% in the study by Jahnsen et al. [7]. Studies that included adults with CP who had learning disabilities reported lower percentages for living independently and having children [39].

A significant number of adults with CP in our study reported having difficulties with daily activities and social participation, especially in mobility, housing, recreation and employment. Similar results were found in other studies [1,5,17,39]. In contrast, adolescents and young adults with CP aged from 16 to 20 years [25] experienced fewer difficulties in daily activities and social participation than our sample, which might be due to a large percentage of unilaterally involved persons, their younger age and the increased likelihood that they still live with their parents.

The results of our study add to the evidence that self-efficacy may contribute to better social participation. This is in accordance with the study by Bent et al. [16] of young adults with a disability, including CP. They found no differences in self-efficacy rates between disabled and non-disabled young adults. Also, in our study, scores for general self-efficacy are comparable to the reference group, except for the higher scores on persisting in the face of adversity. It is possible that persons with CP may have learned to persevere to reach their goals, because of their lifelong disability with barriers.

In addition to participation, this study focused on self-perceived HRQoL, addressing health-related aspects and their perceived impact on functioning. Adults with bilateral CP perceived a low HRQoL for physical functions but not for mental functions. This is in line with other studies that used the SF-36 [7,8,13]. Women with CP had a high mental HRQoL compared with the reference population. Participants with better gross motor functioning were more likely to have a higher physical HRQoL. Livingston et al. [40] reported that the gross motor functioning of children and adolescents with CP was positively correlated with physical functions and physical well-being, but did not consistently correlate with psychosocial well-being. Participants who work hard to achieve their goal (resulting in a higher score on the self-efficacy Effort subscale) were more likely to have a higher physical and mental HRQoL.

Our model for mental HRQoL explained only 16% of the variance in outcome, which is low. This is not surprising, because mental HRQoL could be influenced by different factors that were not included in this study, such as personality, coping styles and social support. The association between self-efficacy and HRQoL is in accordance with the theory of Bandura [14]. He assumed that perceived functioning may be controlled more by beliefs of capability than by the degree of physical impairment, because the latter is not consistent with a patient's perceived quality of life.

Study limitations

A limitation of this study was the low response rate. Only a few adults with CP were currently following a rehabilitation programme (5%).Therefore we had to use a broad approach to include adults with bilateral CP by also searching the historical registers of rehabilitation centres and actively approaching the patients' association to recruit participants. Non-response was due mainly to a lack of up-to-date contact information for potential participants. Furthermore, the burden of the research, including extensive measurements of physical activity and fitness for other parts of the larger study [18], especially in combination with their physical disability, the patient's lack of time available to participate, or lack of interest in the study, e.g. because of a history of extensive medical treatments, were reasons for non-response. Our sample was comparable for gender and level of education to the subgroup of adults with bilateral CP without severe cognitive impairment included in the study of Van der Dussen et al. [39]. In the latter study the sample was checked to be a nearly complete incidence cohort for the region of The Hague, which is part of the recruitment area of the present study [39]. Therefore we assumed the results to be representative for the subpopulation of adults with bilateral CP and normal intelligence. In our opinion, studying subpopulations is a prerequisite for the adequate investigation of functioning and associated factors, thus handling the heterogeneity that is a characteristic of CP. Comparison with other studies in this field is limited because of different selection criteria (e.g., age, subtypes of CP), use of different measurement instruments, and cultural and economic differences (e.g., different levels of unemployment in different regions). The lack of an age-matched control group was resolved by using Dutch reference data.

Implications for treatment

In conclusion, a significant number of adults with spastic bilateral CP without severe cognitive impairment encounter difficulties in daily activities and social participation. Adults with CP perceived a relatively low HRQoL for physical functions, but not for mental functions. Optimising participation in this age group is desirable, especially for mobility, housing, recreation and employment. As self-efficacy was associated with social participation and the Effort subscale with physical and mental HRQoL, psychosocial interventions to improve self-efficacy may enhance both the objective and self-perceived levels of functioning in adults with CP.

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7

Everyday physical activity and community participation of adults with unilateral cerebral palsy

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ABSTRACT

Objective To assess the level and potential determinants of everyday physical activity, and participation in various life areas of adults with unilateral cerebral palsy (CP) in comparison with healthy subjects.

Methods In a cross-sectional study everyday physical activity was measured (Activity Monitor) in 16 adults with CP, aged 28 (SD 3) years, and 16 age/gender matched healthy volunteers, aged 29 (SD 3) years. Participation was assessed by means of validated questionnaires. Age, gender, body fat (skinfold thickness), muscle tone (Ashworth Scale), functional level and participation were assessed as potential determinants of everyday physical activity.

Results In adults with CP mean duration of dynamic activities during a day (10.6 [SD 3.5]%) was comparable to healthy subjects (11.2 [SD 4]%) ($p = 0.66$). In most life areas the level of participation was comparable for both groups, although adults with CP spent more time on non-intensive leisure activities. Participation in sports appeared to be a determinant of everyday physical activity in both groups.

Conclusion The results indicate that the levels of everyday physical activity and community participation in adults with unilateral CP are comparable to levels in healthy comparison subjects.

INTRODUCTION

The Dutch health care system provides many services for children with cerebral palsy (CP). For adults with CP, however, less specific services are available even though the clinical and scientific experience is that adults with CP do report progressive impairments and functional loss [1-7]. These complaints include musculoskeletal problems (e.g., loss of muscle function, increase of joint contractures, pain) [6,8], which often coincide with progressive limitations in everyday physical activities (e.g., walking) [6,7] and in participation in various life areas (e.g., absence from work). Limitations in everyday physical activity can lead to a hypoactive or sedentary lifestyle [9,10], which can aggravate physical complaints, limit participation, have a negative effect on the quality of life, and may lead to the development of cardiovascular diseases [11-14].

To gain more insight in this assumed mechanism of hypoactivity and related problems in adults with CP, we investigated the factors that may contribute to it. In this study we focussed on different aspects of functioning of adults with CP, addressing both level of everyday physical activity and community-related aspects of functioning. Physical activity refers to mobility-related activities, i.e. moving oneself and maintaining and changing postures [15-18]. Community participation refers to a person's involvement in various life situations, defined according to the International Classification of Functioning, Disability and Health (ICF) [19].

There are few reports addressing the level of everyday physical activity in persons with CP. Van den Berg-Emons et al. compared activity levels between 10 children with spastic diplegia and 10 healthy comparison subjects using the doubly-labelled water method and a respiration chamber (total energy expenditure and sleeping metabolic rate, respectively); they found that the children with CP were severely hypoactive [20]. Bandini et al. demonstrated hypoactivity in adolescent wheelchair users with CP, but not in ambulatory patients with CP using doubly-labelled water and a ventilation hood [21].

Although the health status of adults with CP has been described [1,22,23], information on the level of participation of adults with CP in various life areas is still scarce. Andersson et al. and Jahnsen et al. investigated demographic aspects, social situation (education, employment, marital status) and experiences with physiotherapy and physical activity in adult populations with different types of CP [4,24]. Functional capabilities were more extensively studied by Murphy et al. and Bottos et al. [2,5].

This study aimed to assess the level of everyday physical activity and participation of adults with unilateral CP in various life areas compared with healthy matched subjects. In addition, potential determinants of everyday physical activity in adults with unilateral CP and healthy comparison subjects were explored.

We hypothesised that differences would occur between adults with unilateral CP and healthy subjects regarding participation in some life domains, but not with respect to the level of everyday physical activity. This assumption was based on clinical experience and on a study in ambulatory adolescents with CP [21].

METHODS

Recruitment

Sixteen ambulatory adults with unilateral CP aged 25 to 35 years participated in the study (Table 7.1). The subjects were recruited from Sophia Rehabilitation Centres in the Dutch cities of The Hague, Delft and Vlaardingen (south-west region of The Netherlands). Exclusion criteria were: evidence of ataxia or dyskinesia, not being able to walk without help or a walking aid (to avoid misdetection of walking with the Activity Monitor), wheelchair users, other impairments that affect physical activity (e.g., rheumatism, heart failure), a psychiatric diagnosis, behavioural problems or comprehension difficulties which may hamper the assessment.

Sixteen healthy comparison subjects, matched for gender and age (± 3 years), were recruited from the same region via, e.g., relatives or acquaintances of the patients.

In the CP group, 16 of 29 subjects found eligible for the study (55%) agreed to participate. The main reason for not participating was due to the 48-hour measurement with the Activity Monitor (e.g., not being able to shower). There were no important differences in characteristics of participants and non-participants.

The Medical Ethics Committee of Reinier De Graaf Hospital in Delft has approved the study. All subjects were informed about the study and written consent was obtained.

Physical, cognitive and functional characteristics

Body composition and muscle tone

After measuring height while standing and weight on a scale (Seca, Germany) the body mass index (BMI, kg/m^2) was calculated. Thickness of four skinfolds (biceps, triceps, subscapular and supra-iliac) was measured twice with a Harpenden calliper (British Indicators, Burgess Hill, UK); the mean of the two measurements was used for analysis. In the CP group this measurement was performed on the non-hemiplegic side and in the comparison group on the non-dominant side. The percentage of body fat was estimated according to the method of Durnin and Wormsely [25]. Obesity was defined as a percentage body fat > 25% for men and > 32% for women [26].

Muscle tone was estimated by the Ashworth scale on the upper and lower extremities while lying supine. Reliability and validity of the Ashworth Scale are moderate [27,28]. For data analysis, muscle tone (Ashworth scale) was categorised as hypotone, normotone or hypertone.

Cognitive and functional level

The highest completed level of formal education was assessed in both groups to obtain an impression of the cognitive level. The functional level was assessed in adults with CP only, not in the comparison subjects. From a semi-structured interview, based on the different items of the Functional Independence Measure (FIM), scores were derived ranging from 1 (total assistance required) to 7 (complete independence). The FIM includes 18 items to assess functioning in basic physical and cognitive abilities, grouped in a motor dimension (maximum subtotal score 91) and a cognitive dimension (maximum subtotal score 35). The FIM has been found valid and reliable across a wide variety of settings, raters and patients, including patients with CP [29-31].

Outcome measures

Everyday physical activity

The level of everyday physical activity was measured with an Activity Monitor (AM; Temec Instruments BV, Kerkrade, The Netherlands). The AM is based on long-term ambulatory monitoring of signals from body-fixed accelerometers and provides information on duration, rate and moment of occurrence of activities associated with mobility (lying, sitting, standing, walking, cycling, wheelchair-driving, general [non-cyclic] movement) and transitions between postures. Furthermore, information on the variability of the acceleration signal (motility) can be obtained, which is related to the intensity of body-segment movements. In previous studies validity and reliability of the AM has been shown to quantify activities associated with mobility and to detect differences in everyday physical activity between groups [18,32].

The AM consists of four accelerometers (sensors), a portable data recorder worn in a padded bag around the waist, and a computer with analysis programs. Four IC-3031 uniaxial piezo-resistive accelerometers were used (size: 1x1x1 cm) in the standard configuration: one sensor on each thigh and two sensors on the sternum. During standing, the sensors on the thigh and one of the trunk sensors are sensitive in anterior-posterior direction; the other trunk sensor is sensitive in longitudinal direction. Accelerometer signals were stored digitally on a PCMCIA flash card with a sampling frequency of 32 Hz.

After the measurement, data were downloaded onto a computer for analysis by the Kinematic Analysis part of the Vitaport Software [33]. A detailed description of the activity detection procedure can be found elsewhere [9,18,34].

In patients and comparison subjects, a 48-h measurement with the AM was performed during two randomly selected consecutive weekdays during spring or summer. Instrumentation of the AM took place at home or at work to avoid interference with normal activity patterns. To avoid measurement bias, the principles of the AM were explained to the subjects after the measurements. All subjects agreed with this procedure. Data acquired with the AM were calculated per day (24-h period) and averaged over two measurement days. The measurement in one healthy woman was for only 24 hours, she stopped wearing the AM for cosmetic reasons. These results were included in the analysis.

The following variables were assessed as a percentage of a 24-h period: duration of dynamic activities (composite measure: walking, cycling and general movement), and duration of static activities (composite measure: standing, lying and sitting). General movement refers to non-cyclic movements, e.g., moving in the kitchen during cooking. Other variables assessed over a 24-h period were: number of transitions (with and without transitions while lying); number of walking periods longer than 10 seconds, mean body motility (representing intensity of everyday physical activity; motility is expressed in g, 1 g = 9.81 m/s^2) and motility during walking (representing walking speed).

In addition, satisfaction with everyday physical activity was assessed on a vertical Visual Analogue Scale (VAS) of 10 centimetres. The lowest end of the vertical (0 cm) represented no satisfaction, the upper end (10 cm) the best possible satisfaction with everyday physical activity. Visual analogue rating scales have been found to yield reliable and valid data [35].

Participation in various life areas

Sociodemographic data collected addressed housing situation and current employment status. The Dutch version of the domain Occupation of the Craig Handicap Assessment and Reporting Technique (CHART), called the Utrecht Activity List [36,37], and the Community Integration Questionnaire (CIQ) [38,39] were used to quantify an individual's participation in productive activities as well as in home, family and social life. Additional information on the type, duration and frequency of sports activities was obtained with a questionnaire designed specifically for this study. All participants completed the questionnaires in the presence of a researcher. The CHART is a questionnaire that measures deviation from roles generally fulfilled by able-bodied persons [36,37]. The domain Occupation of this questionnaire assesses productive activities, including work, school, household, home maintenance; and leisure activities, addressing volunteer work, sports (club activities) and other recreational activities. Time spent on those activities was recorded in hours per week, rounding up to the nearest hour. The score regarding Occupation was calculated according to the CHART procedure, weighting twice the scores on the items work, school, household and home maintenance. All other items were weighted as one. In this study, we were specifically interested in the level of mobility-related activities. To obtain insight in low-intensive activities, a question was added concerning the time spent on watching television and reading; this question was not included in the CHART score.

The CIQ consists of 15 questions comprising three scales: home integration, social integration and productive activities, (e.g., work, school and volunteer activities). The scoring method of Willer et al. was used. The total CIQ score ranges from 0 to 29, with a higher score indicating a higher level of integration [38,39].

The CHART and the CIQ have high test retest reliability and for both instruments evidence for validity was demonstrated [39,40]. Although the CHART was originally tested on healthy subjects and patients with spinal cord injury, and the CIQ on healthy subjects and patients with acquired brain injury, the questionnaires were considered to be valid to differentiate between adults with CP and healthy subjects.

Potential determinants of everyday physical activity

Several potential determinants of everyday physical activity were explored, both for persons with CP and for healthy comparison subjects. The studied determinants concerned physical characteristics, cognitive and functional level (the latter only in persons with CP), level of participation, and satisfaction with everyday physical activity.

Statistical analysis

Differences between adults with CP and healthy comparison subjects were tested with the Student's *t*-test for independent samples and by means of the χ^2 test for categorical variables. Relationships between parameters were calculated with Spearman rho correlation coefficients (R_s) for parameters on the ordinal level, and Pearson correlation coefficients (R_p) for the interval level. Analysis was performed using the Statistical Package of Social Sciences (SPSS, version 10.0). A probability (p) value of less than or equal to 0.05 was considered as statistically significant, p-values > 0.05 and ≤ 0.10 were indicated as a trend.

RESULTS

Physical, cognitive and functional characteristics

Both study groups consisted of seven men and nine women. Physical characteristics are shown in Table 7.1. In the CP group, eight subjects had a right-sided hemiplegia and eight subjects had a left-sided hemiplegia. Seven subjects had increased muscle tone, six had no increase in muscle tone and one subject had a decreased muscle tone. Two subjects did not want to be examined for muscle tone.

Subjects with CP had a significantly lower body mass than healthy subjects ($p = 0.04$). Men with CP had a lower percentage of body fat than healthy comparison subjects ($p = 0.006$). According to the criteria used, six women with CP (38%), 7 healthy women (44%) and 1 healthy man (6%) were obese.

The subjects' educational and functional levels are reported in Table 7.2. None of the eligible subjects with CP were excluded from the study based on the exclusion criteria regarding limitations in comprehension abilities or ambulation. Four subjects with CP had attended a school for the multiple handicapped (a special form of education for children with a physical handicap combined with cognitive deficits). All other subjects had completed some form of secondary education (see Table 7.2). Adults with CP showed a lower level of education than comparison subjects at a level of $\alpha = 0.10$ ($p = 0.06$).

The adults with CP had a high degree of independence in performing functional activities. The mean score on the motor dimension according to the FIM was 89.1 (SD 3.1) (maximum score: 91) and the mean cognitive score 33.1 (SD 4.4) (maximum score: 35). The total functional score of 122.1 (SD 7) was close to the maximum score of 126 (Table 7.2).

Table 7.1 Physical characteristics of the patients with unilateral cerebral palsy (CP) and their healthy comparison subjects

Characteristics [a]	Patients with CP ($n = 16$)	Healthy subjects ($n = 16$)	p-value
Age (years)	28 (3)	29 (3)	0.90
Gender (number)	7 men, 9 women	7 men, 9 women	1.0
Hemiplegic side (number)	8 right, 8 left		
Muscle tone (number) [b]	1 hypotone, 6 normotone, 7 hypertone		
Height (m)	1.73 (0.08)	1.75 (0.11)	0.44
Body mass (kg)	70.1 (9.7)	77.8 (10.9)	0.04*
Body Mass Index (BMI)	23.7 (3.8)	25.3 (2.4)	0.16
Sum of four skinfolds	63 (31.6)	74.5 (25.5)	0.27
Body fat percentage based on skinfolds (%)	25.6 (9.8)	29.1 (7.1)	0.25
Body fat percentage based on skinfolds (%) in men	16.2 (3.9)	22.2 (2.8)	0.006*
Body fat percentage based on skinfolds (%) in women	32.9 (5.8)	34.5 (3.9)	0.49

[a] Data are mean (SD) or otherwise indicated.
[b] Missing data for $n = 2$.
* Significant difference at level of $\alpha \leq 0.05$.

Everyday physical activity

In both groups no differences were found in the duration of dynamic activities (composite measure: walking, cycling and general movement) between the first and second measurement day. The mean duration of dynamic activities in the CP group was 10.4 (SD 3.1)% on the first day and 10.9 (SD 4.4)% on the second day ($p = 0.55$). In the comparison group this was 11.2 (SD 3.5)% and 11.2 (SD 5.4)%, respectively ($p = 0.98$). Therefore, results of the AM were averaged over the two days. In both groups some persons did and some did not perform sports activities during the measurement days.

Mean duration of dynamic activities during a day was similar in both groups: 10.6 (SD 3.5)% in adults with CP and 11.2 (SD 4.0)% in healthy comparison subjects ($p = 0.66$) (Table 7.3). This corresponds with 2 hours and 32 minutes per day in the persons with CP, and 2 hours and 41 minutes per day in healthy comparison subjects.

Similarly, for other aspects of everyday physical activity measured with the AM, no significant differences were found between the two groups (Table 7.3).

Table 7.2 Patient characteristics: cognitive and functional level and sociodemographic data

Characteristics [a]	Patients with CP ($n = 16$)	Healthy subjects ($n = 16$)	p-value
Level of education			
School for learning disabilities	4	0	0.06
Junior general education and junior vocational education	5	2	
Secondary general education and advanced vocational education	5	10	
Higher educational institutions and university	2	4	
Functional level based on the FIM [b]			
Subscale scores (maximum score)	mean (SD)		
Self-care (42)	41.0 (1.8)		
Sphincter control (14)	13.7 (1.3)		
Transfers (21)	20.8 (0.7)		
Locomotion (14)	13.4 (0.6)		
Motor dimension (91)	89.1 (3.1)		
Communication (14)	13.6 (2.2)		
Social cognition (21)	20.1 (2.2)		
Cognitive dimension (35)	33.1 (4.4)		
Total score (126)	122.1 (7.2)		
Sociodemographic data			
Living situation			
Alone	5	2	0.003*
With parents	5	0	
With partner	4	11	
With others (family, students)	0	3	
Institution	2	na	
Children			
Children	2	6	
Household			
Alone	5	6	
With others	9	10	
Not performed	1	0	
Current employment			
Fulltime employed (≥ 36 hours/week)	6	12	0.07
Part-time employed	8	4	
Unemployed	2	0	

[a] Frequency distribution is given or otherwise indicated.
[b] Missing data for $n = 1$.
* Significant difference at a level of $\alpha \leq 0.05$.
na: not applicable; FIM: Functional Independence Measure.

Figure 7.1 shows the dynamic and static activities specified per activity in percentage a day. For adults with CP time spent on these activities corresponds to 8 hours and 23 minutes lying, 8 hours and 41 minutes sitting, 4 hours and 13 minutes standing, 2 hours and 13 minutes walking and 18 minutes general movement a day.

In both groups, six subjects had cycled during the measurement days; the mean duration was 1.7 minutes a day in the CP group and 8 minutes in the comparison group ($p = 0.05$). There were no significant differences for any of the other activities.

Satisfaction with the level of everyday physical activity was comparable between adults with CP and comparison subjects. On a VAS (0–10) the adults with CP ($n = 15$) had a mean score of 7.2 (SD 2.6) and healthy comparison subjects ($n = 16$) of 7.7 (SD 2.0) ($p = 0.53$).

Participation in various life areas

Table 7.2 presents sociodemographic characteristics of the two groups. In contrast to their age-matched comparison subjects, five adults with CP lived with their parents, and two persons in assisted community housing ($p = 0.003$). Relatively few persons with CP lived with a partner, and only two of them had children. Except for one person with CP, all persons did their own housekeeping, either alone or with another person.

Table 7.3 Mean duration (SD) of dynamic and static activities, number of transitions, walking periods > 10 seconds and motility during a day as measured with the activity monitor

	Patients with CP ($n = 16$)	Healthy comparison subjects ($n = 16$)	*p*-value
Duration dynamic activities [a] % of a day (SD)	10.6 (3.5)	11.2 (4)	0.66
range (% a day)	[5.6 – 16.9]	[5.5 – 21.6]	
Duration static activities [b] % of a day (SD)	89.4 (3.5)	88.8 (4)	0.66
range (% a day)	[83.2 – 94.5]	[78.4 – 94.5]	
Number of transitions	159 (70)	188 (34)	0.15
Number of transitions minus transitions while lying	146 (67)	169 (30)	0.21
Number of walking periods > 10 seconds	211 (70)	215 (65)	0.86
Mean motility (g) [c]	0.037 (0.014)	0.037 (0.012)	0.93
Mean motility during walking (g) [d]	0.25 (0.081)	0.24 (0.085)	0.68

[a] Composite measure of walking (including running and walking stairs), cycling and general movement.
[b] Composite measure of lying, sitting and standing.
[c] Mean motility represents intensity of everyday physical activity, 1 g = 9.81 m/s^2.
[d] Motility during walking represents walking speed, 1 g = 9.81 m/s^2.

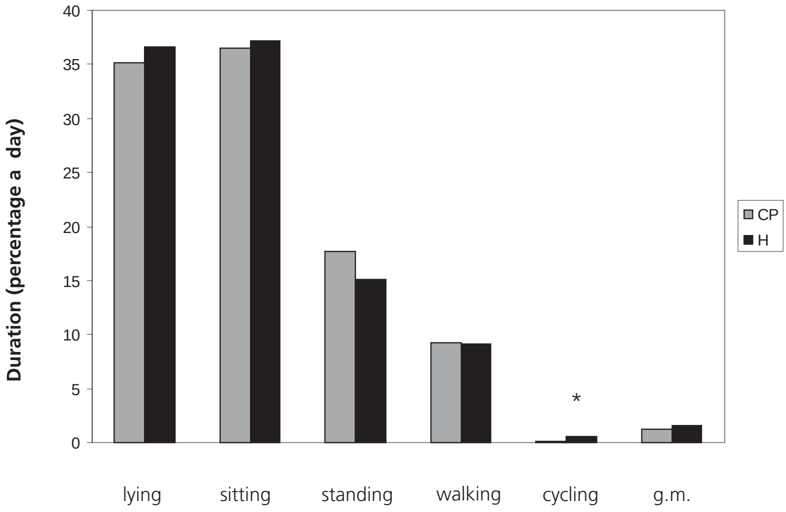

Figure 7.1 Mean duration of dynamic and static activities specified per activity in percentage a day in patients with cerebral palsy (CP) and healthy comparison subjects (H). * significant difference at a level of $\alpha \leq 0.05$; g.m.: general movement.

All subjects in this study, except two persons with CP, worked (Table 7.2). Compared to healthy subjects more persons with CP tended to work part-time or were unemployed, ($p = 0.07$). For the time spent at work, no significant difference was found between those with CP and healthy comparison subjects (Table 7.4), $p = 0.12$.

All subjects travelled almost every day; 14 persons with CP and 13 healthy subjects used a car or public transportation.

Based on the CHART items, adults with CP tended to spend more time on activities like reading and watching television ($p = 0.06$) and other non-intensive leisure activities such as needlecraft ($p = 0.02$) (Table 7.4). Subjects with CP less often took part in volunteer work than comparison subjects ($p = 0.02$); this also applied to this item of the CIQ ($p = 0.006$).

A broad range was found for the total CHART occupation scores of both adults with CP (63–253 points) and healthy subjects (62–168 points). According to Whiteneck et al., a total CHART occupation score of less than 100 was regarded as an occupational handicap [37]. In the present study this applies to 10 adults with CP and 3 healthy subjects.

The level of community integration (including home integration, social integration, and productive activities), as measured with the CIQ was comparable in both groups (Table 7.3). Further analysis showed that 5 subjects with CP who lived on their own had a higher CIQ total score than the 9 CP subjects who lived with a partner, parents or others ($p = 0.04$).

Table 7.4 Participation in productive and leisure activities, community integration and sports

	Patients with CP	Healthy subjects	*p*-value
Productive and leisure activities based on CHART domain Occupation (hours a week) [a]			
Work	28.2 (16.3)	37.8 (17.2)	0.12
School	2.3 (3.9)	2 (5.9)	0.89
Household	17.3 (30.4)	19 (17.6)	0.85
Jobs around the house	1.1 (2.0)	1.3 (1.7)	0.71
Volunteer activities	0.06 (0.3)	0.9 (1.2)	0.02*
Sports	1.6 (1.7)	2.4 (2.4)	0.29
Leisure activities in clubs (no sports)	0.8 (1.6)	1.1 (3.1)	0.72
Other leisure activities (e.g., needlecraft)	5.5 (5.3)	1.7 (2.8)	0.02*
Watching television and reading [b]	16.8 (8.3)	11.8 (5.9)	0.06
Subscale scores			
Productive activities (work, school, household)	97.6 (48.9)	120.3 (29.9)	0.13
Leisure activities [c]	8.0 (6.0)	6.1 (5.5)	0.36
CHART-Occupation score [c]	105.6 (46.7)	126.4 (29.4)	0.14
Community integration [a]			
Subscale scores (maximum score)			
Home Integration (10)	5.8 (2.4)	5.8 (2.1)	0.94
Social Integration (12)	9.0 (2.4)	9.3 (1.9)	0.68
Productive activities (7)	5.7 (1.2)	6.0 (0.63)	0.36
Total score (29)	20.4 (4.0)	21.1 (2.9)	0.58
Sports			
Total number of sporters	11	12	
> 1 sport per person	4	5	
Frequency (times a week)			
0	5	4	
1	5	6	
2 to 3	5	4	
4	1	2	
Duration (hours a week)			
< 0.5	5	4	
0.5 to 3	9	7	
> 3	2	5	

[a] Data are mean (SD) scores.

[b] This an extra category, not included in the CHART scores.

[c] Time spent on reading was not included.

* Significant difference at a level of $\alpha \leq 0.05$.

Of those with CP, 11 took part in sports and 4 performed more than one sport, compared with 12 and 5 subjects, respectively, in the comparison group (Table 7.4). No significant differences were found in duration and frequency of sports between persons with CP and comparison subjects. Relatively many persons with CP were active in swimming and fitness and relatively few were involved in running, ball games and tennis.

Potential determinants of the level of everyday physical activity

Table 7.5 presents an overview of relationships found between determinants and duration of everyday physical activity.

In subjects with CP, a correlation was found between frequency and duration of sport sessions a week and mean duration of dynamic activities a day as assessed with the AM ($R_s = 0.61$, $p = 0.01$; $R_s = 0.50$, $p = 0.05$, respectively). This relationship is shown in Figure 7.2, which gives the interquartile ranges of scores on duration of dynamic activities for three subgroups of adults with CP, addressing non-sporters, persons sporting once, or sporting twice or more in a week. This figure shows that 50% of adults with CP sporting once or at least twice a week were 10.0 to 13.0% or 9.4 to 14.4% of a day dynamically active, as compared to 6.3 to 9.0% of dynamic activities in 50% of the non-sporters. This relationship was confirmed by the correlation found between hours a week spent on sports as measured on the CHART item and mean duration of dynamic activities ($R_p = 0.60$, $p = 0.01$).

Table 7.5 Determinants of everyday physical activity

Determinants	Duration of dynamic activities	
	Patients with CP	Healthy subjects
	R	*R*
Frequency of sports	0.61 ($p = 0.01$)	n.s.
Duration of sports	0.50 ($p = 0.05$)	n.s.
Time spent on sports (CHART)	0.60 ($p = 0.01$)	0.53 ($p = 0.04$)
Satisfaction with level of activity	-0.66 ($p = 0.007$)	n.s.
	Women with CP	Healthy women
Body Mass Index	n.s.	n.s.
Body fat percentage	n.s.	0.62 ($p = 0.07$)

n.s.: non significant; *R*: correlation coefficient.

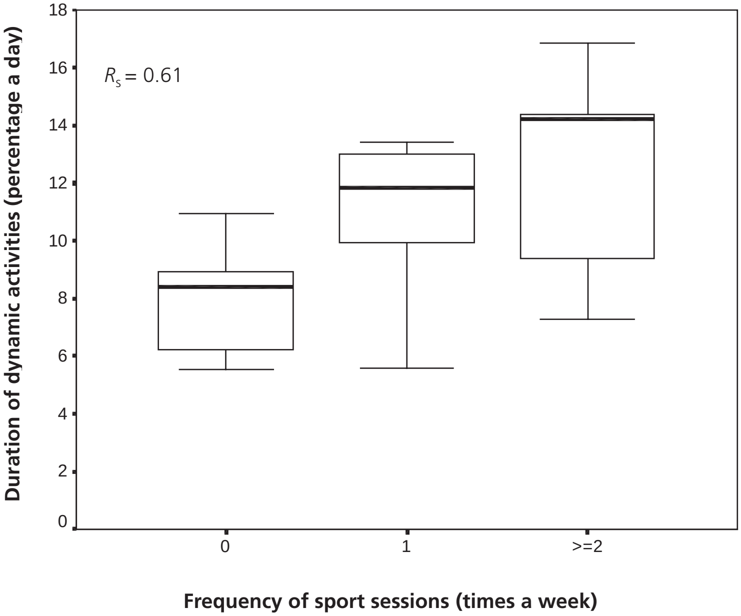

Figure 7.2 Relationship between frequency of sport sessions a week and mean dynamic activity in adults with unilateral CP. Boxplots depict median score (-), interquartile range (white) and minimum and maximum values (whiskers). Subgroups refer to (0) non-sporters, *n* = 5; (1) sporting once a week, *n* = 5; (≥ 2) sporting twice or more a week, *n* = 6.

In subjects with CP a negative correlation was found between satisfaction with the level of everyday physical activity and mean duration of dynamic activities a day (R_s = -0.66, p = 0.007). We found no further evidence for other determinants of everyday physical activity, addressing other physical characteristics, cognitive and functional level, and other aspects of participation.

In healthy subjects only some correlations were found concerning determinants of everyday physical activity (Table 7.4). Time spent on sports (CHART) was related to mean duration of dynamic activities a day (R_p = 0.53, p = 0.04). In healthy women a positive trend was found between percentage body fat and mean duration of dynamic activity a day (R_p = 0.62, p = 0.07).

DISCUSSION

Study sample

In the present study the CP sample had only minor physical impairments; all these subjects were ambulatory, without personal assistance or use of a walking aid. In fact, no persons with CP were excluded from the study because of this criterion. This situation is comparable to a study of Andersson et al., in which all their adult subjects with unilateral CP were able to walk (only one subject was a household ambulator, and one used walking aids) [4].

Our adults with CP had a high functional level according to FIM items, and a relatively low level of education with a broad range. A relatively low level of education was also demonstrated in the study by Andersson et al. amongst Swedish adults with different types of CP, with learning disabilities as an exclusion criterion [4]. Therefore, we believe that our sample was reasonably representative for the adult population with unilateral CP.

Unexpectedly, men with CP had a lower percentage body fat than healthy men. This finding should, however, be interpreted with caution, because the method of calculation we used is based on a method for healthy subjects [25]. However, the method based on the sum of four skinfolds has the same disadvantage.

The percentage of body fat determined from skinfold thickness is probably an underestimation of the real body fat percentage, as demonstrated in children with diplegic CP [41]. Nevertheless, in comparison with a Dutch population in which the prevalence of obesity was 10%, obesity was more prevalent in the women of both groups [42,43].

Everyday physical activity

This is one of the first studies in adults with CP that focused both on everyday physical activity and community participation.

As was expected, adults with unilateral CP did not differ from healthy comparison subjects on the level of everyday physical activity as measured with the AM. In this study all subjects with CP were ambulatory and most of them appeared to have a normotone or a slightly increased muscle tone in their lower extremity at the hemiplegic side, which is not likely to limit mobility-related activities. Of relevance is that the motility during walking, which represents walking speed, was comparable in both groups.

The level of everyday physical activity as measured with the AM in the comparison group is in agreement with measurements with the AM in other healthy comparison subjects of somewhat younger (18 [SD 4] years) [9] or older age (48 [SD 11] years) [44]. Thus, in the present study, the comparison group had a relatively normal level of activity.

The present study demonstrated no hypoactivity in adults with unilateral CP. This is in contrast to more severely affected patients with CP, e.g., Van den Berg-Emons et al. demonstrated hypoactivity in children with diplegic CP, and Bandini et al. in adolescent wheelchair users [20,21].

Participation in various life areas

Contrary to our expectation, adults with unilateral CP had a relatively high level of participation in various life areas. The subjects with CP had a high employment rate. In several reports on adults with CP, persons with hemiplegia had the highest employment rate [2,4]. It should be noted, however, that 50% of our subjects with CP worked part-time.

In the present study the level of community integration (according to the CIQ) did not differ between adults with CP and matched healthy subjects. Regarding participation, only a few differences were found. Adults with CP spent more time on non-intensive leisure activities (e.g., watching television). In a study on disabled children (mostly meningomyelocele and CP), an increased time spent on watching television was found with increasing age [45]. The limited participation in volunteer work by patients with CP might be due to factors other than the health condition. For example, the attitude of the environment towards persons with disabilities might preclude them from volunteer activities.

Both groups travelled by car or public transportation. In this respect, the high functional level of the adults with CP and the easy access to specific transportation services for handicapped people in The Netherlands might play a role. In the survey of Andersson et al. in adults with CP, 33 persons with unilateral CP were able to transport themselves independently whereas 7 subjects experienced difficulties [4].

Few studies amongst adults with CP report on participation in sports activities and physiotherapy [22,24]. Andersson and Mattson supplied some information on frequency of sports activities [4], but not on duration of sports activities. In the present study, no differences were found between adults with unilateral CP and healthy subjects for frequency and duration of sports activities. This might be positively influenced through the stimulation of sports by the Dutch health care system, and the possibilities for handicapped sports and medical fitness in this area of The Netherlands.

Based on the present data it was not possible to classify sports intensity according to criteria of the American College of Sports Medicine Position Stand of 1998 for healthy subjects [46]. From additional information obtained from the questionnaires, however, we estimated that all subjects who took part in sports achieved the adequate duration per session (20–60 minutes per session) and 6 persons in both groups achieved the recommended frequency a week, i.e. 2–3 times per week for muscular strength and endurance, and 3–5 times per week for cardiorespiratory fitness and body composition. The finding that adults with CP were to a lesser extent involved in running, ball games and tennis might be due to their impaired skills regarding co-ordination and speed. This is comparable to a study of Turk et al. who also demonstrated a higher frequency of participation in swimming and walking than in basketball and tennis by both adult men and women with CP [22].

It is difficult to achieve a more detailed comparison of limitations in participation between other studies and ours. This is due to, e.g., our small sample size, a lack of subdivisions between different types of CP or inclusion of other congenital diseases in other studies, use of different classifications of activities, and differences in environmental factors (e.g., differences between countries).

Potential determinants of everyday physical activity

Because of the small sample sizes in both groups, the investigation of potential determinants of everyday physical activity was only of an explorative nature. A larger sample size is necessary to perform multivariable analysis and to achieve more generalisable results.

Nevertheless, in the present study we found that participation in sports was associated to the level of everyday physical activity in both patients and healthy subjects. Participation in other life areas, such as work, was not related to everyday physical activity. For work this might possibly be explained because persons with a full-time sedentary job may have a low activity level because of few periods of walking or general movement during working hours.

A study amongst adults with different types of CP indicated that physical activity may contribute to prevent deterioration of functional capabilities, chronic pain and chronic fatigue [24]. Thus, an improvement of participation in sports may contribute to a reduction of secondary conditions due to inactivity in adults with CP.

A limitation of the present study is that secondary conditions associated with CP (such as fatigue or musculoskeletal pain) which could hamper physical activity in subjects with CP [1,4,5] were not measured. As we found no hypoactivity in the participants of this study, however, the role of these secondary conditions as a determinant of physical activity seems limited.

In the present study, age, gender and muscle tone were not associated with the level of everyday physical activity; this could be expected from the small variation in age, and the normotone or slightly increased muscle tone in persons with CP. Similarly, functional level was not an associated factor for everyday physical activity, which is not surprising because of the high degree of independence we found.

We found that relatively many women were obese, and that healthy women who were obese were relatively active during a day. This relationship was in an opposite direction than expected. Furthermore, in healthy women these results were not unequivocal for the different parameters we assessed regarding BMI and body fat content. Therefore, we cannot conclude that in healthy women body mass or fat content were related to everyday physical activity.

Finally, in adults with CP, a negative correlation was found between a person's satisfaction with the level of everyday physical activity and mean duration of dynamic activities a day. Possible explanations might be that persons with CP who function on a higher level of physical activity either perform physically more demanding tasks or set higher personal goals for functioning.

Conclusion

The results of this study indicate that in most life areas adults with unilateral CP participate to a similar degree as healthy persons, except for non-intensive leisure activities and volunteer work. Regarding sports, differences were found only for type of sports activities performed.

Also, the level of everyday physical activity as measured with the AM in adults with unilateral CP is comparable to levels in healthy comparison subjects. Both patients and healthy subjects appeared to be satisfied with their level of everyday physical activity.

Participation in sports might be a determinant of the level of everyday physical activity in unilateral CP. Further research is necessary to evaluate whether practising sports might be recommended to reduce secondary conditions in adults with unilateral CP who have a low level of everyday physical activity.

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8

General discussion

In the studies presented in this thesis, we examined health issues and participation amongst adults with spastic cerebral palsy (CP) without severe cognitive impairment. One sample included adults with bilateral CP aged 25 to 45 years and a second, smaller sample included adults with unilateral CP aged 25 to 35 years. The first study addressed a wider range of topics because we hypothesised that we would find a significant risk on health issues and participation restrictions amongst adults with bilateral CP.

KEY FINDINGS

Adults with spastic bilateral CP had a high prevalence of chronic pain, fatigue and depressive symptoms compared to healthy reference samples. Persons with more severe fatigue experienced more depressive symptoms as well. Reduced levels of aerobic fitness were found in adults with bilateral CP compared to healthy reference samples. The level of everyday physical activity was also reduced, especially amongst those persons with CP who had lower levels of gross motor functioning. Adults with unilateral CP had everyday physical activity levels comparable to those of able-bodied age mates. Although the 10-year fatal cardiovascular (CVD) risk was low amongst participants with bilateral CP, several single CVD risk factors were present, (pre)hypertensive blood pressure levels being the most significant. Higher body fat tended to be related to higher 10-year CVD risk. Lower aerobic fitness levels were moderately associated with higher levels of fatigue in men and with lower self-reported physical activity in women.

With respect to participation, adults with bilateral CP experienced difficulties in several domains, primarily related to mobility, housing and recreation. They perceived a low health-related quality of life (HRQoL) for physical but not mental functions. Amongst adults with unilateral CP, participation was comparable to that of an able-bodied reference sample for most life areas; however, adults with unilateral CP spent more time on non-intensive leisure activities.

For persons with bilateral CP, those with a higher sense of general self-efficacy were more likely to have higher levels of social participation, and persons who expended more effort in completing their goals were more likely to have a higher physical and mental HRQoL. Chronic pain and fatigue were not associated with difficulty in daily activities or social participation.

Subgroup analyses demonstrated that women with bilateral CP had lower levels of aerobic fitness and a lower 10-year fatal CVD risk compared to men. No gender differences were found for other health issues or participation. Those adults with bilateral CP and lower levels of gross motor functioning experienced more frequent depressive symptoms, more difficulty in daily activities and social participation, and perceived a lower physical HRQoL. In both samples, neither age nor educational level was associated with health issues or participation.

In the following paragraphs, limitations and strengths of the studies, as well as the significance of the main findings will be discussed.

STUDY LIMITATIONS AND STRENGTHS

Study samples

We conducted two cross-sectional cohort studies in relatively young adults with spastic CP. Both studies had relatively small sample sizes: 56 adults with bilateral CP, 16 adults with unilateral CP, and 16 able-bodied age mates. Based on a power-analysis, the minimum sample size was set at 50 participants with bilateral CP. However, a larger sample would be preferable to study prevalence and associations. One reason for these sample sizes was the extensive measurements required of participants. Secondly, only a minority of persons with CP in The Netherlands retains contact with a rehabilitation care provider during adulthood. Less than one-third of persons with CP in a Dutch study by Hilberink et al. [1] and 5% of those in the present study sample were receiving concurrent rehabilitation treatment. Consequently, these persons are difficult to trace, due to changing addresses, and may not be inclined to participate in research.

Considering CP-related disability, the low prevalence of CP, and recruitment challenges, we chose reasonable sample sizes which were comparable to those used in other studies of adults with CP [2-6]. A strength of the current study is our broad recruitment strategy, which contrasts with some studies in the USA which recruited participants from amongst those adults currently known at medical centres [3,5,7].

For both studies, we expended considerable effort to reach adults with CP. In addition to registers of adult rehabilitation patients, historical registers of paediatric rehabilitation patients were used to trace adults with CP. Adults with unilateral CP were recruited from the mid-west region of The Netherlands, including cities, small villages and the countryside. For adults with bilateral CP, we recruited participants from 10 rehabilitation centres in the western and central regions of The Netherlands and via the Patient Association BOSK [8]. We covered a large area of the country in an effort to make our results representative for The Netherlands.

Of the invited eligible persons minus excluded persons, 38% of adults with bilateral CP and 55% of adults with unilateral CP participated. This response rate is similar to that observed in surveys targeting persons with comparable diagnoses such as a history of polio [9]. Reasons given for declining were: no reason, burden of the research, and lack of time. Non-response studies showed that there were no important differences in characteristics between participants and non-participants.

CP-related and demographic characteristics of our participants with bilateral CP are comparable to those for a subgroup of adults with bilateral CP without severe cognitive impairment reported in a nearly complete Dutch incidence cohort study from the same geographic region [10]. In consideration of these similarities, we assume our study samples to be sufficiently representative.

The exclusion criteria 1) multi-morbidity with lasting effects on physical activity (unilateral and bilateral CP); and 2) contraindications for progressive maximal cycle ergometry and full dependence on electric wheelchair propulsion (bilateral CP), may have affected levels of aerobic fitness and everyday physical activity, prevalence of health issues, and participation in our studies. However, including persons with multi-morbidity or contraindications may have created a selection bias. Including persons dependent on electric wheelchair propulsion would have resulted in a more heterogeneous study sample. Furthermore, studying this electric wheelchair-dependent group requires other measures. Moreover, selection bias may have occurred when fitter persons participated because of the study title, 'daily functioning and physical fitness'. Persons with severe cognitive impairment were excluded because of challenges in understanding study instructions and questionnaires; they may also experience different life course perspectives on autonomy. Earlier studies show that prevalence of health issues and participation differ amongst persons with CP based on the presence or absence of severe cognitive impairment [11,12]. Ethically, those with severe cognitive impairment were excluded because of their legal inability to grant consent for participation.

Our research focuses on the 25 to 35 year-old and 25 to 45 year-old age bands and extends the longitudinal CP Transition study, which began with participants aged 16 to 20 years old and included follow-up measurements at year two and year four [13-16].

Study design

The cross-sectional study design allows for frequency measurements but is limited in its ability to study cause-effect relationships. Therefore, demonstrated associations are not conclusive, but rather show associations at a particular moment in time and suggest avenues for future research.

We compared our results with various Dutch reference samples. In adults with unilateral CP, able-bodied comparison persons ($n = 16$) were recruited from the same region and matched for gender and age. To study everyday physical activity levels in persons with bilateral CP, reference values for healthy age- and gender-matched subjects were obtained from our own database. An advantage of this method is that these comparison persons were measured by the same procedures.

Except for aerobic fitness (see below), all other study parameters for bilateral CP were compared with reference samples from previous studies in The Netherlands. The primary data sources used were the population-based data from the Monitoring Project on Chronic Disease Risk Factors (MORGEN-project) from the National Institute of Public Health and the Environment (RIVM) [17] and Statline from Statistics Netherlands (CBS) [18]. These data were available for similar age groups and included comparable measurement procedures (e.g., CVD risk factors). Our results for fatigue, depressive symptoms, HRQoL and self-efficacy were compared to Dutch studies using the same questionnaires. We realise that comparison with population-based studies has methodological limitations (e.g., differing sample sizes). However, we prefer comparisons to general populations, in consideration of the relatively high prevalence of chronic pain, fatigue and sedentary behaviour amongst the general population. Finally, health issues were investigated primarily amongst adults with bilateral CP. Adults with unilateral CP were less extensively studied, because fewer health problems were expected in this group.

Strengths of this study include the extensive objective measurements of everyday physical activity, aerobic fitness and CVD risk. These extensive measurements contrast with other studies which frequently used self-reported data. In addition to daily activities and participation, self-perceived functioning was assessed. Furthermore, the research projects fill a gap in the literature on adults with CP. Next to the Norwegian study of Jahnsen and Opheim [19,20] it is one of the first studies to examine a wide range of health and health-related topics in adults with both unilateral and bilateral CP.

Measures

In addition to the study limitations described in Chapters 2 to 7, a few remarks should be made regarding the measures used.

Questionnaires

One strength of the research is that data on health issues and participation were collected via face-to-face interviews (e.g., chronic pain) and by various self-report questionnaires administered in the presence of a trained researcher (e.g., Short Form-36 Health Survey) [21]. It should be noted that, generally, questionnaires are subjective and carry the risk of producing socially desirable answers.

Fatigue is especially difficult to assess, considering its multidimensional construct. The Fatigue Severity Scale (FSS) [22] concerns fatigue severity and impact on daily life; it is, therefore, more related to behavioural consequences of fatigue. In addition, we used the Multidimensional Fatigue Inventory (MFI-20) [23,24] to gain insight into the nature of fatigue.

The questionnaires that measure participation and HRQoL were considered suitable to provide insight into (self-perceived) functioning and to have adequate psychometric properties for application to adults with CP. We switched from the Community Integration Questionnaire (CIQ) [25,26] and the Dutch version of the domain Occupation of the Craig Handicap Assessment and Reporting Technique (CHART) [27,28] in persons with unilateral CP to the Assessment of Life Habits 3.0 (LIFE-H) [29] in persons with bilateral CP because the Dutch language version of the LIFE-H has shown to be reliable [30] and is more frequently used in persons with CP [12-14].

Aerobic fitness

Aerobic fitness was measured during a progressive maximal exercise test on an electronically braked cycle ergometer, based on the McMaster All-Out Progressive Continuous Protocol [31], a gold standard test for persons with CP [32]. According to the averages of peak heart rate, respiratory exchange ratio and perceived exertional score, participants generally reached nearly maximal exercise performance ($VO_{2\ peak}$). $VO_{2\ peak}$ was used as a main outcome measure. It is suggested to express $VO_{2\ peak}$ in normalised values: body mass (mL/kg/min) or fat-free mass (mL/kgFFM/min). We expressed $VO_{2\ peak}$ in L/min throughout all analyses, in consideration of the following arguments. Earlier studies [33], as well as clinical experience and our findings, suggest that body composition in CP may differ from that of able-bodied persons. To date, lower, similar and higher body fatness have been demonstrated in adults with

CP when compared to healthy persons [34]. Secondly, the effect of body mass was expected to be small because cycle ergometry is a non-weight bearing test. Thirdly, analyses with $VO_{2\,peak}$ corrected for body mass (mL/kg/min) produced similar results in our study. Fourthly, estimation of fat-free mass would be based on skinfold measurement using a calculation method developed from a healthy population sample [35]. These may be inappropriate, because skinfold measurements underestimate body fat percentage in children with CP [35,36]. We corrected for gender to address differences in fat-free mass between men and women.

In two studies we investigated aerobic fitness (Chapters 4 and 5). During the course of the study, we switched from using the reference values of Vos [37] to the reference values of Shvartz [38]. Values reported by Vos were measured indirectly with submaximal tests in Dutch sedentary adults. Values reported by Shvartz were measured directly with maximal cycle ergometry in healthy, untrained samples from North American and European countries (including The Netherlands). Similar results were obtained when we compared results using reference values reported by Vos and Shvartz.

Everyday physical activity

Everyday physical activity level was measured using an accelerometry-based activity monitor (AM) during a 48-hour period on two randomly selected consecutive weekdays [39]. It is suggested that monitoring should be performed over three to five days in adults and seven days in children [40] to show habitual physical activity behaviour. However, our aim was not to characterise habitual physical activity, but to provide insight into everyday physical activity levels. Therefore, we consider our two randomly selected weekday monitoring sessions in both persons with CP and able-bodied age-mates justified. This method is supported by White et al. [41], who considered a 24-hour AM-measurement to be of adequate duration to reliably record activities. In the present studies, the chosen method is further supported by the finding that no significant differences were noted in the duration of dynamic activities between the first and second measurement days.

Limitations of AM use are described in detail in Chapters 3 and 7. One strength is that the AM provides detailed information about several aspects of everyday physical activity [42], in contrast to more commonly used step-watch monitors [43] or actometry devices. Secondly, the AM provides objective data in contrast to subjective questionnaires, which have been used in earlier studies on physical activity in adults with CP [44,45] and may overestimate physical activity. Thirdly, the AM is useful for detecting inter-group differences in levels of everyday physical activity [39,46].

Cardiovascular disease risk

Non-fasting blood samples were used to avoid the negative influence of prolonged fasting on outcome measures such as the maximal ergometer test. Assessing non-fasting blood samples may overestimate CVD risk. However, there is growing evidence supporting the use of non-fasting values to assess CVD risk [47,48]. Although Dutch guidelines recommend measuring fasting lipid profiles and glucose, they allow non-fasting values to estimate CVD risk as well [49]. The SCORE [50] steering committee of the European Society of Cardiology (ESC) [personal communication] and the NCEP guidelines [51] allow measurement of non-fasting total cholesterol (TC) and high-density lipoprotein cholesterol (HDL-C) as well.

Statistical analyses

In addition to topics described in Chapters 2, 5 and 6, some discussion of the regression analyses is needed. For multivariable models we selected independent variables based on clinical experience and previous research, and explored associations generated by univariable regression analysis. To accommodate for the small sample size, we used, at most, three independent variables in each multivariable analysis. We presented basic models and extended models with one independent variable added at a time.

The lack of associations demonstrated in some parts of the study may be due to the methods used. The first possible explanation is our use of strict criteria (e.g., severe fatigue instead of fatigue as a dependent variable). Secondly, focusing on the degree of difficulty instead of frequency of daily activities and participation may have obscured true associations. A third possible explanation is our dichotomisation and the chosen cut-off points. However, some variables in our study were not normally distributed and required dichotomisation. Fourthly, other independent variables could have been explored in regression analyses. For example, considering the large proportion of reportedly single persons in the current studies, we could have corrected for having a partner. Studies amongst general populations demonstrate that physical activity is related to the social environment. Lower physical activity levels have been reported previously amongst people who are single, have a lower educational level, or are socially isolated [52].

MAIN FINDINGS

Health issues

Chronic pain was reported by 75% of adult participants with spastic bilateral CP, which is significantly more than the 39% reported in the reference population of the same age [53,54]. Sixty-eight percent had pain lasting longer than one year. Multiple pain sites were significantly more prevalent in adults with bilateral CP compared to the reference sample. Most pain sites were located in the back and lower extremities. These findings underscore the seriousness of pain and are consistent with other studies in adults with CP [1,5,6,19,55].

The present study is one of only a few to address fatigue and depressive symptoms in persons with CP. One out of five adults with bilateral CP was fatigued and a further two out of five were severely fatigued. Both severity of fatigue and nature of fatigue subscale scores were significantly higher compared to Dutch healthy reference samples [56,57]. Opheim and Jahnsen [20] reported similar fatigue severity in Norwegian adults with CP.

Depressive symptoms were demonstrated in one-quarter of adults with bilateral CP, which was significantly more than the reference group [58]. Two other studies investigating depression in a broad age range of persons with CP reported prevalences ranging from 10% to 35% [11,59]. Fatigue and depressive symptoms should be considered as significant health problems in adults with CP.

We measured aerobic fitness in ambulatory adults with bilateral CP. Average aerobic capacity was 77% of reference values, which supports the evidence that aerobic fitness levels are reduced in adults with CP [34,60-62]. During the past few decades, increasing evidence of low aerobic fitness amongst children and adolescents with CP [63,64] further justifies its importance as a treatment goal.

As expected, everyday physical activity levels were reduced in adults with bilateral CP, primarily amongst those with lower levels of gross motor functioning. Duration of average dynamic activities (116 min/day versus 157 min/day) and mean motility (intensity of activity) were both significantly lower compared with able-bodied age-mates. In contrast, adults with unilateral CP had similar durations of dynamic activities as able-bodied age-mates (152 min/day versus 161 min/day). Other aspects of everyday physical activity were also similar between those with unilateral CP and able-bodied age-mates. It should be noted that all adults with unilateral CP were ambulatory and the sample size was small, which limits the generalisability of these results. The inactive lifestyle amongst persons with bilateral CP is consistent with results of studies amongst diplegic children [65,66] and adolescents with various CP types [43,67,68], as measured by a variety of procedures.

In contrast to adults with bilateral CP, persons with unilateral CP with longer durations of dynamic activities appeared to be less satisfied with their level of everyday physical activity. A possible explanation may be that adults with unilateral CP who function at a higher everyday physical activity level intend to have 'normal lives' and set higher goals or perform more demanding tasks.

Amongst those with bilateral CP aged 25 to 45 years, the 10-year fatal CVD risk was low (< 1%). However, several single CVD risk factors were present, suggesting a risk of developing CVD at older age. Apart from the aforementioned reduced levels of aerobic fitness and everyday physical activity, higher blood pressure was demonstrated in persons with CP, including a two-fold higher rate of hypertension in men compared with the reference sample [17]. Obesity was more often reported in men and less often in women compared to the reference groups. A large waist circumference was present in approximately one-fifth of the study sample. In contrast, lipid profiles were more favourable, and those with bilateral CP reported less smoking and alcohol consumption compared to reference samples. Blood glucose levels were normal.

Disentangling health issues

In this section we intend to disentangle the nature and interrelationships of the health issues we studied.

Chronic pain

To our knowledge, the origins of pain have not been studied systematically in adults with CP. Adults with bilateral CP in this study self-reported a variety of assumed causes of chronic pain: muscles or muscle cramps, osteoarthritis, arthritis, tendinitis, bursitis, hip dysplasia, carpal tunnel syndrome and physical overuse. These possible causes may result from neuromotor impairment with spasticity, muscle weakness, musculoskeletal deformities and pathological movement patterns. Considering that most pain sites were located in the back and lower extremities, bilateral distribution and abnormal gait pattern may play a role. Neuropathic pain causes were infrequently reported. Myelopathy and radiculopathy may be expected more often in other CP subtypes (e.g., dystonic) and at older ages. Adults with CP did not offer psychological and personal factors as related to their pain. It should be noted that chronic pain is a complex, multidimensional experience which includes physical, cognitive, emotional, motivational and behavioural components. Origins of chronic pain require further study.

Physical inactivity, reduced aerobic fitness and fatigue

Several explanations are possible for the physical inactivity observed in adults with bilateral CP. The first explanation for physical inactivity may be the motor impairment itself. This hypothesis is supported by the finding that adults with bilateral CP with lower levels of gross motor functioning had lower levels of everyday physical activity. However, no associations with other parameters generally affecting gait, such as spasticity, were demonstrated (Table 8.1). We might have missed an association because we used a global indicator of spasticity (number of spastic muscles in the lower extremity) for participants with bilateral CP. In unilateral CP, muscle tone was not associated with everyday physical activity. To our knowledge, no literature exists regarding the association between spasticity and physical inactivity.

Secondly, an abnormal gait pattern may influence everyday physical activity by decreasing walking efficiency and increasing oxygen consumption [69]. Increased oxygen consumption during everyday physical activities such as walking has been reported in children [70-72] and adults [73] with CP. Increased oxygen consumption during walking, together with reduced levels of aerobic fitness, may cause increased physical strain during walking (oxygen uptake during walking as a percentage of peak aerobic capacity) [70,74]. It is suggested that high levels of physical strain in CP may be a cause of physical inactivity and fatigue [70,74,75]. Our results from another study using the same data set (not included in this thesis) showed a higher level of physical strain during walking at preferred walking speed in bilateral CP compared to able-bodied adults [76]. Furthermore, physical strain during walking at preferred walking speed was inversely related to duration of walking in daily life [76]. These findings add to the evidence that physical strain relates to physical activity and that persons with CP use a large proportion of their metabolic reserve for walking compared with able-bodied persons. We found no evidence for a direct relationship between physical strain during walking and severity of fatigue [76].

Thirdly, earlier studies suggest that fatigue itself could result in less walking and lower levels of physical activity [69-71], as persons attempt to avoid symptoms of fatigue. In Norwegian adults with CP, a lack of physical activity was associated with fatigue in univariable analysis [77]. In the present study, we found no direct association between fatigue and everyday physical activity (Table 8.1). One reason for a lack of demonstrated association (apart from methodological considerations) is the multidimensional nature of fatigue. This hypothesis is underscored by the high scores observed on all scales of the nature of fatigue, including general, physical and mental fatigue, as well as reduction in activities and motivation. Thus far, we have studied relationships with severity of fatigue and not with other fatigue characteristics. Another explanation may be that an imbalance between everyday physical activity and aerobic fitness affects fatigue more than the levels of everyday physical activity and aerobic fitness themselves.

A fourth explanation for physical inactivity in bilateral CP may be the high occurrence of chronic pain and depressive symptoms. However, we have not yet studied relationships between everyday physical activity and chronic pain or depressive symptoms. However, one Norwegian study indicated that adults with CP and a low prevalence of secondary impairments, such as pain, were more frequently physically active [45].

Fifthly, participation, personal factors, and environmental factors may explain the level of everyday physical activity. In an additional study on bilateral CP (not included in this thesis), we found no relationship between everyday physical activity and participation [78]. In unilateral CP, everyday physical activity level was associated with participation in sport (Table 8.1), but not with other areas of participation such as employment. Also, no relationships were found with body fat, cognitive level or functional level in persons with unilateral CP.

Finally, we found little evidence for the assumption that everyday physical activity and aerobic fitness interact in our cross-sectional study (Table 8.1) [79]. In women with bilateral CP, reduced aerobic fitness was moderately associated with lower self-reported physical activity levels. However, this finding was not confirmed by objective physical activity measurements or in men with bilateral CP. In general populations [80] and amongst adults with other chronic conditions [81-83], associations between aerobic fitness and everyday physical activity level are also inconclusive. In the general population, a large part of the variability in fitness is not accounted for by physical activity [80]. In contrast, interrelationships do exist amongst non-ambulatory young adults with myelomeningocele [81] and adults with a recent spinal cord injury [82] who have lower levels of everyday physical activity and aerobic fitness compared to our study sample. These findings suggest that threshold values, below which relationships occur, may play a role.

With respect to reduced aerobic fitness levels, we found that, although the association was not strong, men with lower aerobic fitness levels reported higher levels of fatigue. However, there was no such relationship amongst women with bilateral CP (Table 8.1). Thus, the evidence for direct roles of everyday physical activity and fatigue resulting in reduced levels of aerobic fitness is weak. The findings on physical strain in bilateral CP imply that persons

Table 8.1 Associations studied with aerobic fitness and everyday physical activity level in adults with cerebral palsy

Potential associated factors	Aerobic fitness (bilateral CP)	Everyday physical activity (unilateral and bilateral CP)
Sociodemographic and CP-related factors		
Age	na	-
Gender	+	-
Educational level [a]	na	-
GMFCS	-	+, bilateral CP only
Limb distribution	na	-
Muscle tone [b]	na	-
Physical condition and complaints		
Aerobic fitness ($VO_{2\ peak}$)	na	-
Level of everyday physical activity (AM)	-	na
Self-reported physical activity (PASIPD)	+, women only	na
Satisfaction with level of everyday physical activity (VAS)	na	+, unilateral CP only
Sport (frequency and duration of sessions per week)	na	+, studied in unilateral CP only
Severity of fatigue (FSS)	+, men only	-
10-year fatal CVD risk (SCORE low-risk)	-	-
Blood pressure (sphygmomanometry)	-	-
Body fat (BMI, sum of four skinfold)	na	-, studied in unilateral CP only
Participation and health-related quality of life		
Daily activities (LIFE-H)	-	-, studied in bilateral CP only
Social participation (LIFE-H)	+	-, studied in bilateral CP only
Physical HRQoL (SF-36)	+	-, studied in bilateral CP only
Mental HRQoL (SF-36)	-	-, studied in bilateral CP only

[a] Educational level: Low: no education or elementary school and pre-vocational practical education; Medium: pre-vocational theoretical education and upper secondary vocational education (vocational high school); High: secondary education, higher education and university

[b] Tardieu scale in bilateral CP; Ashworth scale in unilateral CP

CP: Cerebral palsy; AM: Activity monitor; PASIPD: Physical Activity Scale for Individuals with Physical Disabilities; VAS: Visual analogue scale; FSS: Fatigue Severity Scale; SCORE: Systematic Coronary Risk Evaluation; LIFE-H: The Assessment of Life Habits short version 3.0; SF-36: Short Form-36 Health Survey; na: not assessed; +: associated factor; -: no association demonstrated.

who experience high physical strain during walking are likely to walk less than persons with low physical strain during walking. Because physical strain is defined as oxygen consumption as a percentage of peak aerobic capacity, it may be that aerobic fitness is indirectly related to walking duration [76]. In another study on the dataset of persons with bilateral CP, adults with higher aerobic fitness levels seemed to have both less difficulty in social participation and a higher physical HRQoL [78]. Furthermore, factors mentioned in the disentanglement of physical inactivity, such as chronic pain, depressive symptoms and sports participation, may play a role in the demonstrated reduced aerobic fitness levels.

Fatigue and depressive symptoms

In addition to the physical factors discussed in the above paragraph, factors such as muscle weakness, reduced muscle mass or alterations in muscle structure may play a role in CP-related fatigue [69,84,85]. Central fatigue, a decreased voluntary activation of muscles by the nervous system [86], may be present in CP as well.

No difference between physical and mental fatigue has been demonstrated in adults with bilateral CP. Jahnsen et al. found that adults with CP reported significantly more physical, but no more mental fatigue, compared to the general Norwegian population [77]. The role of brain damage in fatigue or depressive symptoms (as occurs in acquired brain injuries) [85] or central sensitisation in fatigue is not yet clear for persons with CP.

Fatigue may be linked to other symptoms as well [85]. In adults with bilateral CP and severe fatigue, 83% suffered from chronic pain and 44% from depressive symptoms. Those with both chronic pain and fatigue had the highest percentage of depressive symptoms (48%). This finding is consistent with the general population, in which the co-occurrence of pain and fatigue is consistently high and depression is more common in persons who report both pain and fatigue compared to those who report either symptom or neither [87]. Although associations between chronic pain and fatigue have previously been demonstrated in adults with CP [5,55,77], in other diagnoses [87,88], and in the general population [87], we could not confirm this with our findings. Contrary to our expectations, chronic pain and depressive symptoms were not related. In addition to methodological factors, this may have resulted from the apparent acceptance of pain by persons with CP as part of their lifelong disability. Furthermore, other aspects of lifelong disability, such as deterioration of body functions, may contribute to the development of depressive symptoms.

Severe fatigue and depressive symptoms occurred frequently in adults with bilateral CP and often in combination. It is unlikely that these findings can be explained solely by the assessment methods, because fatigue (assessed by the FSS) and depressive symptoms (assessed by CES-D) have been shown to be distinct entities in persons with and without chronic diseases [22]. One possible explanation is that symptoms are linked as a result of medication side effects. Medication prescribed for pain and other symptoms in persons with CP (e.g., opioid analgesics, anti-depressants, or anti-epileptics) may produce fatigue as an adverse effect [85,87]. The effect of coping styles, which may play a role in these symptoms, requires further study. Furthermore, symptoms may be reported in parallel, but are not causally linked.

The relatively high prevalence of depressive symptoms in persons with lower gross motor functioning, suggests that the physical disability may coincide with feelings of depression. In fact, these persons may have more accompanying impairments (e.g., affected vision or communication) [89] which may in turn influence mood. The possibility that the more severe brain damage in persons with lower levels of gross motor functioning is a contributing factor is unlikely considering the weak correlations between neuroimaging and the clinical presentation of CP [89].

Cardiovascular disease risk

Higher waist circumference and body mass index (BMI) tended to be related to a higher 10-year fatal CVD risk in adults with bilateral CP. This trend is consistent with results of other studies on body fat and CVD risk in the general population [49,90]. No associations, however, were demonstrated between everyday physical activity or aerobic fitness and the 10-year fatal CVD risk. Considering the high number of adults with bilateral CP who had (pre)hypertensive blood pressure (BP) values, we studied potential associated factors. Mean diastolic BP (DBP), but not mean systolic BP (SBP), was related to waist circumference and BMI; however, no relationships were demonstrated with aerobic fitness and everyday physical activity.

Concluding remarks

In summary, our results suggest that health issues in adults with bilateral CP may be caused by many interacting components. Considering the results of everyday physical activity, we found not much support for a central role of everyday physical activity in developing or aggravating physical complaints as suggested by the cycle of deconditioning described by Durstine [79] and noted in studies of persons with other neurological conditions [91]. Our results provide some support for a role of aerobic fitness in the cycle of deconditioning (Table 8.1), suggesting that adults with bilateral CP may benefit from sport and exercise. However, caution is needed in interpreting these results because of the small sample size, relatively young age of participants, high level of gross motor functioning, and exclusion of persons dependent on electric wheelchair propulsion. Inclusion of persons fully dependent on electric wheelchair propulsion (GMFCS level IV or V) or persons who could not perform cycle ergometry may have produced broader ranges of aerobic fitness and everyday physical activity, as well as possible associations with 10-year CVD risk. Further research on activity patterns (e.g., time spent on sedentary versus high-intensity activities such as cycling or running) and energy expenditure may be useful to deepen our understanding of everyday physical activity and its assumed relationships.

Impact on participation in life areas

Adults with CP in both samples had a high degree of independence in performing basic functional activities, as measured by the Functional Independence Measure (FIM) [92]. With respect to indicators of participation, however, participants had disadvantaged positions for living with a partner and having children, as compared to reference groups of similar ages [18]. Adults with CP from other Western countries also score low on these indicators [77,93,94]. Furthermore, adults with CP tended to lag behind able-bodied peers in employment levels (including sheltered employment). Of adults with bilateral CP, 54% were competitively employed and 14% had sheltered employment, compared to 77% of persons with paid employment in the general population [18]. Compared to able-bodied persons, more adults with unilateral CP tended to be unemployed or to work part-time. However, employment rates in the current studies seemed high compared to other European countries, which showed rates of only 33% to 47% [2,77,93,95]. Overall, a significant number of adults with bilateral CP encountered difficulties in complex daily activities (mobility 77%, housing 71%, personal care 45%) and in social participation (recreation 60%, employment 44%), despite the use of assistive devices, adaptations or human assistance [29]. In adults with unilateral CP, participation in most life areas was similar to that of able-bodied age-mates, with the exception of having more non-intensive leisure activities and less volunteer work. They performed other types of sports such as swimming and fitness, and were involved to a lesser extent with running, ball games and tennis.

Based on mean scores for most LIFE-H categories [29] and the CHART occupation score [28], one may conclude that adults with CP function relatively well on average. The variance around the mean scores or the dichotomised ‘having difficulty’ score from the LIFE-H presents a different picture, one in which a substantial number of persons with bilateral CP are restricted in activities and social participation. Adults with unilateral CP had a mean score of 106 on the CHART; however, an occupational handicap (score of less than 100) was present in 10 of 16 adults.

Our findings regarding daily activities and participation are consistent with other studies in CP [93,94,96,97]. However, compared to Dutch adolescents and young adults with CP and normal intelligence [13], adults with bilateral CP perceived more difficulties in daily activities and social participation. In addition to younger age and inclusion of all CP types in the study of Donkervoort et al. [13], fewer difficulties in activities and participation may be associated with living at home with parents.

With respect to self-perceived health and functioning, adults with bilateral CP had a significantly lower physical but not mental HRQoL compared to the general population. These HRQoL findings are similar to those found in other studies of adults with CP [44,77,98]. General self-efficacy [99] was similar amongst adults with bilateral CP and reference controls; however, adults with CP were significantly more persistent in the face of adversity. Perhaps persons with CP generally learn to persevere to reach their goals because of their lifelong disability. General self-efficacy was associated with social participation. This finding adds to the existing evidence in young adults with several disabilities that self-efficacy may contribute to better participation [100]. In addition, persons who work hard to achieve their goals are more likely to have a higher HRQoL.

As mentioned previously, adults with bilateral CP and higher levels of aerobic fitness, seemed to have less difficulty in social participation and have higher physical HRQoL [78]; we found no support for an association between everyday physical activity and these outcomes [78]. Pain and fatigue are assumed to be associated with daily functioning in CP; however, the effect of these symptoms on daily functioning varies across studies [5,19,55,77,101]. In our study, we could not demonstrate an association between chronic pain, severe fatigue or depressive symptoms and daily activities or social participation.

Subgroups

Gender

In contrast to the general population and persons with other chronic conditions, in which women suffer from pain, fatigue and depressive symptoms more frequently than men [87], we found no clear gender differences for these symptoms amongst persons with CP. Nor did vitality and mental health show significant relationships with gender. These findings contrast with those of a Norwegian study which reported that women with CP experience a higher frequency and intensity of pain [19,20] and a higher total and physical fatigue [77]; however, this study included all CP subtypes and a wider age range.

The finding that aerobic fitness and 10-year risk of fatal CVD were lower in women with bilateral CP is consistent with findings in general populations [49] and other studies of aerobic fitness in CP [102]. Gender was not associated with everyday physical activity level in the present study or amongst adolescents with CP in previously published studies [43,66]. Participation and HRQoL were unaffected by gender in persons with CP, except for mobility, which was significantly lower for women than men with bilateral CP. The lack of association with gender may be due to the overriding influence of the physical disability on health.

GMFCS level

Subgroup analysis with GMFCS level showed that gross motor functioning is a relevant correlate in adults with bilateral CP. A significantly lower everyday physical activity level was demonstrated amongst adults with bilateral CP at the GMFCS level III/IV compared to GMFCS level I. In adults with unilateral CP, a rather homogeneous sample in this respect, no significant differences for GMFCS level were found. These results are similar to those reported for children with CP [66]. Higher gross motor functioning was positively associated with daily activities and social participation in persons with bilateral CP. Studies of children with CP [96] and Dutch young adults with CP [13] showed similar results. Participants with better gross motor functioning were more likely to perceive a higher physical HRQoL. This association supports the findings of Rosenbaum et al. [103] in adolescents with CP. Furthermore, persons at the GMFCS level III/IV perceived more depressive symptoms.

For other health issues (e.g., aerobic fitness, 10-year fatal CVD risk) no associations were found with gross motor functioning. The lack of an association between gross motor functioning and fatigue is difficult to interpret, as severity of fatigue is often related to disease severity. It is possible that studying a larger sample may show an association between fatigue severity and GMFCS level, considering the nearly significant *p*-value of 0.08 demonstrated in this study. Chronic pain prevalence did not differ by GMFCS levels. The lack of an association may be explained by lower everyday physical activity levels or wheelchair use by those with lower gross motor functioning, sparing them from pain-inducing activities. However, some previous studies report higher levels of chronic pain in persons with CP who are non-ambulatory and quadriplegic [3,5].

Conclusion

Various health issues and participation limitations were observed in the relatively young adults with bilateral CP; all of them had at least one additional health problem or participation restriction. Interestingly, these health issues and limitations co-occur with low physical HRQoL. These findings underscore the severity of additional consequences of bilateral CP. Gross motor functioning appears to be an important correlate in adults with bilateral CP. Relevant modifiable factors in bilateral CP seem aerobic fitness, body fat and self-efficacy.

Adults with unilateral CP had normal levels of everyday physical activity; these levels were inversely related to the participants' subjective satisfaction with everyday physical activity level. Considering their minimal limitations in daily activities and participation, these persons function at a relatively high level.

CLINICAL IMPLICATIONS

During the past decade, CP at adult age has gained increasing research attention. Considering the low prevalence of CP and the fact that few adults with CP are in contact with rehabilitation care, it is unlikely that adult health care workers have extensive treatment experience or up-to-date scientific knowledge to help this patient group [1]. Therefore, it is important to educate health care workers in several areas. Firstly, diagnosis of health issues within the complex disorder of CP is needed. For example, health care workers may be unaware of the high occurrence of depressive symptoms or may not recognise mood problems in combination with a variety of other complaints. Secondly, it is important to improve providers' knowledge of modifiable factors and behavioural change. Thirdly, prevention and intervention programmes are critical. The following paragraphs will elaborate on these topics.

Considering the childhood onset of CP, there may be a central role for rehabilitation medicine in treatment and research. The complexity of the disorder and additional consequences require rehabilitation centres with specialised rehabilitation physicians and therapists. However, less technical aspects of screening, prevention and treatment (e.g., CVD risk assessment), could be addressed in primary care. The high occurrence of health problems and participation restrictions encountered in this lifelong disability requires that a lifespan care approach and self-determination be important aspects of treatment. Applying these approaches from a young age onwards may benefit not only persons with CP but also society by reducing long-term treatment and costs. To address high health care costs, group training, modular programmes and internet-based programmes may be cost-effective and feasible options. These methods may have other advantages such as positive group interactions and avoidance of travelling by persons with disabilities.

Health issues

The variety of self-reported causes of chronic pain requires a thorough examination of physical causes. However, other chronic pain components (e.g., behavioural) must be considered as well. The multidimensional character of fatigue emphasises the importance of a thorough clinical interview and physical examination.

Currently, a variety of treatment options exist to address pain, fatigue and depressive symptoms. In the present study, more than half of adults with bilateral CP received care from a physical therapist. Medications for pain and spasticity were used in less than 25% of adults with bilateral CP, and few used antidepressants or underwent psychological treatment. Our findings support previous studies in adults with CP that showed high use of physical therapy and low use of psychological interventions [1,45,104], combined with pain which is undertreated [1,7,104]. It is likely that adults with CP are also undertreated with respect to other available therapies such as spasticity treatment and surgical procedures.

Thus far, few studies have investigated the effect of treatment in adults with CP. Jensen et al. [104] demonstrated that pain medication and most physical therapy treatments are rated as providing pain relief, but no treatment was consistently associated with large decreases in pain. Despite the variety of physical therapy treatments received by our adult participants with bilateral CP, there remained a high prevalence of chronic pain and fatigue. This unmet need suggests that preventive measures and more effective interventions are necessary to reduce these symptoms in CP. Future studies should aim to assess the effectiveness of physical therapy.

The relatively young age of our study sample and the growing evidence of pain in children with CP [105,106] underscore the necessity to begin treatment at an early age. The high prevalence of health issues, specifically the co-occurrence of pain, fatigue and depressive symptoms, warrant a holistic approach and treatment of symptom clusters. Rehabilitation teams are experienced in some of these approaches, such as in treatment of chronic pain or acquired brain injury. For instance, cognitive behavioural therapy (CBT) [107], psychomotor therapy (PMT) [108] and balancing of activity and rest may be helpful for persons with CP.

CBT has demonstrated efficacy in many conditions, including pain, fatigue, depression and weight loss amongst both children and adults. The evidence for CBT is still emerging in various conditions [107]. There is some evidence that adults with CP could benefit from the CBT model of chronic pain [4,109]. Pain-coping strategies appeared to be associated with changes in functioning over time in a sample of adults with CP-related chronic pain. Coping strategies that target the coping responses of catastrophising, rest and task persistence appeared to be more closely linked to pain interference and depressive symptoms compared to other strategies [4,109]. CBT may also benefit those experiencing fatigue or weight problems, and help persons cope with the consequences of their CP disability and augment medical self-management. In developing a CBT intervention for adults with CP, these factors may improve efficacy: 1) addressing various health issues and health-related problems; 2) considering accompanying impairments (e.g., cognitive impairments); and 3) systematic and repeated training of coping strategies [4].

PMT may be a treatment option for some adults with CP, considering the co-occurrence of physical and psychological complaints and disturbed body perception [110]. PMT is a holistic approach to improving both physical and psychological well-being and enhancing physical fitness in patients suffering from various conditions [108]. Thus far, most evidence for its efficacy has been shown in mental illness [108,111]. In rehabilitation care, clinical experience is growing in children with CP [112] and adults with chronic musculoskeletal pain.

Most Dutch rehabilitation programmes encourage balancing of activity and rest. The hypothesis behind this treatment is that both overuse and hypoactivity [79] should be avoided to prevent symptom aggravation and physical deconditioning. To our knowledge, neither measures for balancing physical capacity and load nor the effectiveness of these approaches have been studied in adults with CP.

With respect to everyday physical activity and aerobic fitness, there are some clinical implications to consider. Adults with bilateral CP, both those with high and low GMFCS levels, did not achieve the minimum physical activity recommendations for healthy persons according to American College of Sports Medicine (ACSM) [113]. Periods of continuous dynamic activity for of at least 10 minutes were achieved by only 39% of participants. It should be discussed whether ACSM criteria for the general population are applicable to persons with CP, considering their motor impairment.

A previous study amongst adults with different CP types suggests that physical activity may prevent chronic pain, chronic fatigue, and deterioration of functional capabilities [19,77]. Thus far, we found no evidence that improving everyday physical activity directly benefits health issues. However, our results do suggest a role for aerobic fitness in interrupting the cycle of deconditioning and underscore the benefits of regular sports and exercise participation for adults with bilateral CP. Aerobic fitness training may break through the deconditioning cycle and improve health and participation in those with bilateral CP. A previous study of exercise in children with CP showed favourable effects on aerobic fitness [63]. An advantage of aerobic fitness training is that it can be performed in relatively safety through aquatic training, stationary cycling or tailored fitness.

Although the 10-year CVD risk was low, several single biological and lifestyle-related risk factors were present in our relatively young study sample. In addition to the aforementioned health issues, CVD risk factors seemed to be underdiagnosed amongst persons with bilateral CP. Our findings highlight the importance of screening for CVD risk factors, specifically blood pressure and body fatness, and to start preventive strategies from young adulthood onwards [114]. Considering that persons with higher waist circumferences or BMIs tended to have a higher 10-year CVD risk, some support was found for weight management in bilateral CP. In this respect, the recommendations of the Dutch Guideline Cardiovascular Risk Management could be applied to persons with CP [49]. In addition, the amount of sedentary time and its relationship with CVD risk should be studied.

Participation

Optimising participation is desirable, especially for adults with bilateral CP. Some of the aforementioned approaches, such as aerobic fitness training [63] and CBT [107], have been shown to produce favourable effects on participation and quality of life for persons with CP [4,63,109]. Most adult CP participation restrictions could be regularly treated by outpatient neuro-rehabilitation teams, which have extensive experience in this area. However, only a minority of adults with CP are in contact with rehabilitation care. Adults with CP could use online information to identify and contact with specialised rehabilitation care programmes. At Rijndam Rehabilitation Centre, we recently developed a CP informational website [115]. The Patient Association BOSK [8] also provides counselling information.

A more fruitful approach may be to prevent participation restrictions in CP during adulthood. Those developing prevention programmes should create programmes that: 1) start at a young age; 2) provide continuity with rehabilitation care after discharge from paediatric rehabilitation; and 3) train self-determination skills to optimise autonomy in several life areas.

Over the past several years, increasing numbers of young people have been referred from paediatric to adult rehabilitation care and transitional care programmes have been developed. In 2006, the first 'young adult team' was established at the outpatient adult clinics of Erasmus MC and Rijndam Rehabilitation Centre [115,116]. The young adult team is a transitional service for young people, aged 16 to 25 years, with childhood-onset disabilities. In addition to regular rehabilitation programmes, a specialised modular rehabilitation programme is provided. The aims of the young adult team are: 1) continuity of rehabilitation care from childhood to adulthood; 2) monitoring and treatment of the medical condition; 3) education on the long-term consequences and advice on prevention; 4) achievement of life skills to optimise autonomy; and 5) improvement of daily living activities and participation in several life areas.

Self-determination and self-efficacy towards behavioural change are key factors to optimise autonomy and participation. The present study indicated that higher self-efficacy may enhance participation and HRQoL in adults with CP. Self-efficacy can be learned through psychosocial interventions such as mentoring and training in self-determination skills such as decisionmaking, problem-solving and assertiveness [117]. Our findings underscore the self-efficacy theory of Bandura, that developing self-determination is important to gain maximal personal competence and succeed in achieving life goals [118,119]. The young adult team, including the modular rehabilitation programme, incorporates this theory into the treatment approach [116]. The module 'manage your life' provides training in self-determination and self-efficacy. Using these various treatment options, we aim to facilitate functioning of persons with CP across the lifespan.

DIRECTIONS FOR FUTURE RESEARCH

Amongst the implications of our findings is the need for additional studies to gain further insight into underlying mechanisms and potential treatment targets.

Current dataset

Fortunately, the current dataset for persons with bilateral CP contains more data than presented in this thesis and allows for easy access to study future research questions. Thus far, we have studied two components of health-related physical fitness: aerobic fitness and body composition. Additionally, we wish to analyse neuromuscular fitness (e.g., muscular strength) and its relationships with factors such as fatigue, everyday physical activity and CVD risk. This knowledge could contribute to our understanding of whether strength training interventions to improve physical fitness or everyday physical activity could be explored in adults with CP.

Regarding the data on everyday physical activity, further analysis of sedentary time and high-intensity activities is of interest. Previous studies have shown that greater sedentary time is associated with an increased risk of fatal and non-fatal CVD [120].

Studying sports participation and potential associated factors (e.g., social support, enjoyment, aerobic fitness and everyday physical activity) could provide insight into factors contributing to sports participation as well as relevant associations with aerobic fitness and everyday physical activity. This topic is of special interest because rehabilitation services are well-suited to provide advice and sports training. Other factors potentially associated with everyday physical activity and aerobic fitness such as pain, nature of fatigue and depressive symptoms could be studied as well.

Finally, self-reported exacerbating and relieving factors of pain and possible causes of self-perceived fatigue and fitness could be explored to use the experiences of adults with CP as source of information. Similarly, studies of the use of medication, allied health care, and adaptive aids could reveal targets for treatment studies.

Future projects

Considering the complex health issues experienced by persons with CP, future research should primarily focus on the level of impairment. In the section on Clinical Implications, we introduced treatment options with points of relevance. Additionally, it would be worthwhile to develop an intervention study tailored for adults with CP, based on CBT and secondly PMT. A challenge of this type of study would be to form efficient and effective modular programmes and group therapies. Self-determination and self-efficacy are incorporated in the young adult team programme and could be added to the adult intervention as well. Furthermore, we plan to study the effectiveness of the module 'manage your life'.

One frequently asked question is whether physical therapy should be continued long-term in adults with CP. Because of the high use of physical therapy and the scarcity of evidence addressing methods and supporting its effectiveness, further research is warranted. We are awaiting the results of the Dutch Learn to Move national research programme on the effects and underlying mechanisms of age-specific treatments for children and young adults with CP [121]. The part of this multi-centre, randomised controlled trial studying 16 to 24 year-olds is being performed by researchers at Erasmus MC and Rijndam Rehabilitation Centre. The study, which is partly based on results described in this thesis [122], intends to permanently increase physical activity and fitness, as well as examine effects on fatigue, participation and HRQoL.

Some of the health issues we studied require further investigation. The aetiology of chronic pain and fatigue requires further research to reveal potential targets for prevention and treatment. Of special interest would be examining dose-response relationships of physical activity and aerobic fitness, as well as measuring the (im)balance between physical capacity and load. Regarding the inconclusive results on body fat and risk for osteoporosis in persons with neurological disabilities, body composition could be more thoroughly investigated. Furthermore, nutrition and energy expenditure in relation to physical activity could be studied in adults with CP.

The current studies were cross-sectional. Longitudinal studies would be of interest to investigate how health issues evolve and to gain insight into health risks at older ages. For example, a larger follow-up cohort of adults could enrol adults aged 45 to 65 years, including persons with unilateral CP to determine whether they are at risk of developing long-term health issues.

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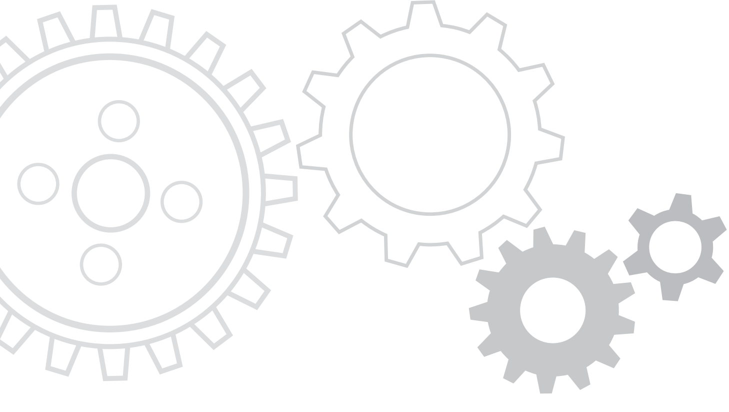
9

Summary
Samenvatting



SUMMARY

Cerebral palsy (CP) is defined as a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to nonprogressive disturbances that occurred in the developing foetal or infant brain. The motor disorders of CP are often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour, by epilepsy, and by secondary musculoskeletal problems.

The permanent and complex nature of CP and the risk of developing secondary conditions imply that persons with CP will face health issues and participation restrictions throughout life. Therefore, it is necessary to study each life phase. However, in contrast to CP in childhood, CP in adulthood is scarcely studied. The main aim of this thesis is to improve our insight into health issues and participation amongst adults with spastic CP, and to identify targets for prevention and treatment of both health problems and participation restrictions.

We conducted two cross-sectional studies amongst adults with spastic CP without severe cognitive impairment. Fifty-six adults with bilateral CP aged 25 to 45 years and 16 persons with unilateral CP aged 25 to 35 years as well as 16 able-bodied age-mates were enrolled in the studies. The first study addressed more topics because we hypothesised a higher risk of health issues and participation restrictions in adults with bilateral CP. Below, a brief summary of the main results per chapter of this thesis is presented.

In Chapter 2 we examined the prevalence and co-occurrence of chronic pain, fatigue and depressive symptoms in adults with spastic bilateral CP and explored associations of chronic pain and fatigue with depressive symptoms and daily functioning. We found that adults with bilateral CP had a significantly higher prevalence of chronic pain (75% versus 39%) and depressive symptoms (25% versus 12%) than Dutch healthy reference samples. One out of five participants was fatigued and a further two out of five were severely fatigued. Both the severity and nature of fatigue scores were significantly higher in persons with bilateral CP than in Dutch healthy reference samples. Chronic pain and severe fatigue co-occurred in 34% and in combination with depressive symptoms in 16% of the participants. Severity of fatigue was associated with depressive symptoms. Chronic pain and fatigue were not associated with limitations in daily activities and social participation. These findings suggest that relatively young adults with spastic bilateral CP are severely affected by these symptoms, in addition to their spastic paresis. More research is needed on the aetiology of chronic pain, fatigue and depressive symptoms to develop preventive measures and interventions to effectively reduce these symptoms in adults with CP.

In Chapter 3 we investigated the level of everyday physical activity in adults with spastic bilateral CP compared with able-bodied age-mates. We explored potential associations with personal and CP-related characteristics, to identify subgroups at increased risk of inactivity. The level of everyday physical activity was measured objectively using an accelerometry-based Activity Monitor. Duration of average dynamic activities (116 min/day versus 157 min/day) and intensity of activity were both significantly lower compared with able-bodied age-mates. Periods of at least 10 minutes of continuous dynamic activity occurred in only 39% of participants, meaning that most adults with bilateral CP did not achieve the minimum physical activity recommendations for healthy persons according to the American College of Sports Medicine. Gross motor functioning was significantly associated with level of everyday physical activity. We found no association between mean duration of dynamic activities and satisfaction with level of everyday physical activity. The results suggest that adults with spastic bilateral CP, especially those with low-level gross motor functioning, are at risk of an inactive lifestyle. Several factors might play a role in the observed physical inactivity, which are disentangled in Chapter 8.

In addition to everyday physical activity, we assessed one of the components of health-related physical fitness: aerobic fitness. Chapter 4 addresses aerobic fitness and its relationships with everyday physical activity and fatigue in ambulatory adults with spastic bilateral CP. On average, the aerobic capacity of adults with bilateral CP measured by a progressive maximal aerobic cycle test was 77% of Dutch reference values. A lower level of aerobic fitness was related to lower self-reported levels of everyday physical activity in women and to higher levels of fatigue in men. No other relationships were demonstrated between these parameters. The results suggest that ambulatory adults with bilateral CP have low levels of aerobic fitness. Thus far, we found little evidence for relationships between the level of aerobic fitness and everyday physical activity or fatigue. However, the small sample size, high level of gross motor functioning of participants or other factors described in the study limitations might contribute to this lack of associations. Further research involving other measures and/or a different sample is required.

Cardiovascular disease (CVD) risk factors and 10-year clustered risk of a fatal cardiovascular event in adults with bilateral CP were examined in Chapter 5. As body fat, aerobic fitness and everyday physical activity are modifiable factors from an exercise perspective, we explored associations between the 10-year risk and these factors.

The 10-year fatal CVD risk was low (< 1%) in spastic bilateral CP. However, several single CVD risk factors were present in these relatively young adults. Apart from the aforementioned reduced levels of everyday physical activity and aerobic fitness, higher blood pressure levels were demonstrated in persons with CP, including a twofold higher hypertension rate in men compared to a reference sample. Also, obesity based on body mass index (BMI) was more often present in men with bilateral CP; a large waist circumference was found in about one-fifth of the study sample. Lipid profile, smoking and alcohol consumption were more favourable amongst adults with bilateral CP than in a reference sample, and blood glucose levels were normal. Higher body fat tended to be related to higher 10-year risk. No associations, however, were demonstrated between physical activity or aerobic fitness and the 10-year fatal CVD risk. These results suggest a risk of developing CVD in adults with spastic bilateral CP at older age. They also underline the importance of screening for CVD risk factors, specifically monitoring blood pressure, and starting preventive strategies from young adulthood onwards. Considering that persons with a higher waist circumference or BMI tended to have a higher 10-year CVD risk, some support was found for weight management.

In Chapter 6 we examined participation and health-related quality of life (HRQoL) in adults with spastic bilateral CP, and explored associations with self-efficacy. At least 60% of the study sample had difficulty in mobility, recreation and housing, and 44% in personal care and employment. Adults with bilateral CP perceived a low HRQoL for physical functions, but not for mental functions. General self-efficacy was similar in adults with bilateral CP and a reference group; however, adults with CP were significantly more persistent in the face of adversity. Study participants with higher general self-efficacy were more likely to have higher social participation, and persons who expended effort in completing their goals were more likely to have higher physical and mental HRQoL. Adults with bilateral CP and lower levels of gross motor functioning had more difficulty in participation and a lower physical HRQoL. These findings suggest that a significant number of adults with bilateral CP encounter difficulties in activities and participation and perceive a low HRQoL for physical functions. As general self-efficacy was associated with social participation, and the effort subscale with HRQoL, a psychosocial intervention to improve self-efficacy may enhance both the objective and self-perceived levels of functioning in adults with CP.

In Chapter 7, we focus on adults with spastic unilateral CP. The level of everyday physical activity and participation of adults with spastic unilateral CP, as compared to able-bodied age-mates, was investigated. In addition, potentially associated factors of everyday physical activity and satisfaction with level of everyday physical activity were explored. The adults with unilateral CP had a similar duration of dynamic activities as able-bodied age-mates (152 min/day versus 161 min/day); other aspects of everyday physical activity were also comparable. Persons with unilateral CP with longer durations of dynamic activities appeared to be less satisfied with their level of everyday physical activity. In most life areas the level of participation was comparable for both groups, although adults with CP spent more time on non-intensive leisure activities. Participation in sports was associated with everyday physical activity in both groups. The results indicate that the levels of everyday physical activity and participation in adults with unilateral CP are comparable to the levels in healthy matched persons. It should be noted that all adults with unilateral CP were ambulatory.

Chapter 8 addresses the main findings and relates them to previous studies. Furthermore, study limitations and strengths are discussed, including sample size, response rate and methods. Strengths of the study are the objective measurements and the wide range of health-related topics examined in adults with both unilateral and bilateral CP.

In conclusion, the studies presented in this thesis show that various health issues and participation limitations were observed in the relatively young adults with bilateral CP. All of the persons with bilateral CP had at least one additional health problem or participation restriction. These findings coincide with a low perceived physical HRQoL. The study examined modifiable factors in this patient group. We found not much support for a central role of everyday physical activity in developing or aggravating complaints as suggested by the cycle of deconditioning of Durstine. However, aerobic fitness, body fat and self-efficacy seemed relevant factors in bilateral CP, and could be modifiable.

In adults with unilateral CP, normal levels of everyday physical activity were demonstrated, which was inversely related to the person's subjective satisfaction with the level of everyday physical activity. Considering their minimal limitations in daily activities and participation, they function relatively well.

The main directions for future research are to disentangle the underlying mechanisms of health issues, and to refine targets for prevention and treatment. Relevant topics for intervention studies are discussed in Chapter 8 as well. Amongst the clinical implications of the studies are the need for adequate diagnosing of health issues in adults with CP, and the importance of education of health care workers. The high (co)-occurrence of symptoms and participation restrictions in this lifelong disability indicates the need for a holistic and lifespan care approach. The results on modifiable factors might be used to develop preventive measures and intervention programmes. An example is training in self-determination skills to improve self-efficacy. Training in aerobic fitness is discussed, as this could break through the cycle of deconditioning and benefit both health and participation in bilateral CP. However, more insight into the role of everyday physical activity, physical fitness and sports in countering health problems is required. Knowledge of treatment in other diagnoses or medical specialities, such as cognitive behavioural therapy and balancing of physical capacity and load, might be used in the treatment of symptom clusters. With these and other discussed implications, we aim to contribute to a lifespan care approach for persons with CP.

SAMENVATTING

Cerebrale parese

Cerebrale parese (CP) wordt gedefinieerd als een groep persisterende houdings- of bewegingsstoornissen die leiden tot beperkingen in activiteiten door een niet-progressieve beschadiging van de hersenen tijdens hun ontwikkeling. De aandoening gaat vaak gepaard met stoornissen in waarnemen, communicatie, cognitieve functies (bijvoorbeeld geheugen), gedrag, epilepsie en met secundaire problemen van het houdings- en bewegingsapparaat.

Vóór 2000 werd CP in Nederland 'infantiele encephalopathie' genoemd of benoemd op basis van klinische kenmerken, zoals spasticiteit of hemiparese. In Nederland is de prevalentie van CP circa 2 per 1.000 levend geboren kinderen. CP is niet te genezen. De levensverwachting van het type CP onderzocht in dit proefschrift is vergelijkbaar met die van de algemene bevolking.

Verschillende oorzaken kunnen leiden tot hersenbeschadiging bij CP, zoals doorbloedingsstoornissen van de hersenen bij te vroeg geboren kinderen. De ernst en de uitgebreidheid van CP kunnen variëren. In de eerste levensjaren kan de uitingsvorm van CP nog veranderen. Later kunnen onder invloed van de groei, door de bewegingsstoornis met spasticiteit en de afwijkende biomechanische eigenschappen (bijvoorbeeld spierverkorting) veranderingen optreden in het bewegingsapparaat. Voorbeelden zijn contractuurvorming en luxatie van gewrichten.

Bewegingsstoornissen bij CP zijn op verschillende manieren in te delen. De indeling op basis van lokalisatie betreft een eenzijdige stoornis (unilateraal) of tweezijdige stoornis (bilateraal). Naar de aard van de stoornis onderscheidt men spastische, atactische en dyskinetische CP. Dit proefschrift richt zich op de grootste groep bewegingsstoornissen: spastische CP (85%), zowel bij unilaterale als bilaterale beelden. Het type CP wordt geclassificeerd volgens de 'Reference and Training Manual of the Surveillance of CP in Europe (SCPE)', de ernst van CP op basis van de 'Gross Motor Function Classification System (GMFCS)'.

Aanleiding en doel

Naar kinderen met CP is meer onderzoek gedaan dan naar volwassenen met CP. Onze eigen klinische ervaring, en die van patiëntenvereniging BOSK, is echter dat volwassenen met CP zich geregeld melden met diverse klachten, bijvoorbeeld moeheid of pijn. Vaak komen deze klachten voor in combinatie met een achteruitgang van het functioneren. Voorbeelden zijn problemen met dagelijkse activiteiten uitvoeren (waaronder lopen) en werken.

Hoofddoel van dit proefschrift is inzicht geven in gezondheidsklachten en participatieproblemen bij volwassenen met spastische CP. Dit om aangrijpingspunten te vinden voor preventie én behandeling van klachten en om participatie te optimaliseren. Met dit proefschrift willen wij bijdragen aan een levensloopbenadering voor mensen met CP.

De gezondheidsklachten die wij onderzoeken houden verband met de houdings- of bewegingsstoornissen (bijvoorbeeld pijn) en observaties uit de praktijk zoals moeheid. Voorafgaand aan ons onderzoek veronderstelden wij dat CP en de klachten als gevolg daarvan gepaard kunnen gaan met minder lichamelijke activiteit en een lage fysieke fitheid. En dat de klachten een negatief effect kunnen hebben op het uitvoeren van dagelijkse activiteiten en participatie. Vanuit de algemene bevolking is bekend dat lichamelijke inactiviteit en een lage fitheid de kwaliteit van leven negatief kunnen beïnvloeden en de kans op het ontwikkelen van hart- en vaatziekten en diabetes vergroten. Daarnaast kunnen beide leiden tot een negatieve spiraal met deconditioneren zoals is beschreven door Durstine.

Onderzoeksopzet en methoden

In dit proefschrift beschrijven we twee dwarsdoorsnedeonderzoeken naar mensen met spastische CP zonder ernstige cognitieve stoornissen. Eén onderzoek is uitgevoerd bij 56 mensen tussen de 25 en 45 jaar met bilaterale CP (hoofdstuk 2-6). Een ander kleinschaliger onderzoek heeft plaatsgevonden bij zestien personen met unilaterale CP tussen de 25 en 35 jaar en zestien vergelijkbare leeftijdgenoten zonder beperking (hoofdstuk 7). In het onderzoek naar mensen met spastische bilaterale CP zijn meer onderwerpen aan bod gekomen, omdat we verwachtten dat zij meer risico hebben op gezondheidsklachten en beperkingen in het dagelijks functioneren.

De ‘International Classification of Functioning, Disability and Health’ (ICF) van de Wereldgezondheidsorganisatie (WHO) is gebruikt om gezondheid en gezondheidsgerelateerde domeinen te classificeren. Het proefschrift is onderverdeeld in ‘gezondheidsklachten’ en ‘participatie’. Onder participatie wordt het deelnemen aan het maatschappelijk leven verstaan. De mate van dagelijkse lichamelijke activiteit (bewegen) is ingedeeld bij gezondheidsklachten. Alle andere activiteiten, zoals huishoudelijke activiteiten, hebben we beschreven in de context van participatie.

Dagelijkse lichamelijke activiteit is gedurende 48 uur ambulant geregistreerd met een activiteitenmonitor. Deze activiteitenmonitor meet met versnellingssensoren verschillende aspecten (o.a. duur en frequentie) van lichamelijke activiteit gerelateerd aan beweging zoals lopen, fietsen en zitten.

Aërobe fitheid, een component van fysieke fitheid, hebben we gemeten met een maximale aërobe inspanningstest (fietsergometrie). Lichamelijk onderzoek (inclusief bloedafname) is verricht naar spiertonus, lichaamsvet en andere risicofactoren voor hart- en vaatziekten. Overige gezondheidsfactoren, participatie en demografische kenmerken hebben we geïnventariseerd met vragenlijsten en semigestructureerde interviews.

De resultaten bij mensen met CP hebben we vergeleken met referentiegroepen uit de algemene bevolking, omdat gezondheidsklachten zoals chronische pijn, moeheid en lichamelijke inactiviteit ook vaak voorkomen in de algemene bevolking.

In hoofdstuk 2 onderzoeken we chronische pijn, moeheid en depressieve symptomen bij volwassenen met bilaterale CP. We bestuderen relaties tussen deze symptomen onderling en de impact ervan op dagelijkse activiteiten en participatie.

Hoofdstuk 3 heeft tot doel het kwantitatief en objectief meten van de dagelijkse lichamelijke activiteit bij volwassenen met bilaterale CP en om de resultaten te vergelijken met die van leeftijdgenoten zonder lichamelijke beperking. Om subgroepen met een verhoogd risico op een inactieve leefstijl te onderscheiden onderzoeken we relaties van persoonsgebonden en CP-gerelateerde factoren met dagelijkse lichamelijke activiteit.

In hoofdstuk 4 bepalen we de aërobe fitheid bij volwassenen met bilaterale CP die kunnen lopen. Tevens onderzoeken we de relaties tussen aërobe fitheid, dagelijkse lichamelijke activiteit en moeheid.

In hoofdstuk 5 onderzoeken we bij volwassenen met bilaterale CP biologische en leefstijlgerelateerde risicofactoren voor hart- en vaatziekten en schatten we het geclusterde tienjaarsrisico op sterfte door hart- en vaatziekten. Relaties tussen het tienjaarsrisico en lichaamsvet, aërobe fitheid en dagelijkse lichamelijke activiteit onderzoeken we om inzicht te krijgen in factoren die het risico op hart- en vaatziekten positief kunnen beïnvloeden.

In hoofdstuk 6 bestuderen we participatie, gezondheidsgerelateerde kwaliteit van leven en self-efficacy (geloof in eigen kunnen) bij volwassenen met bilaterale CP. We onderzoeken of er een verband is tussen participatie of gezondheidsgerelateerde kwaliteit van leven enerzijds en opleidingsniveau, motorisch functioneren (GMFCS) of self-efficacy anderzijds.

In hoofdstuk 7 onderzoeken we de mate van dagelijkse lichamelijke activiteit en participatie bij volwassenen met unilaterale CP. De resultaten vergelijken we met die van vergelijkbare leeftijdgenoten zonder beperking. Daarnaast onderzoeken we relaties tussen dagelijkse lichamelijke activiteit enerzijds en fysieke kenmerken, cognitief niveau, participatie en tevredenheid met de dagelijkse lichamelijke activiteit anderzijds.

Belangrijkste bevindingen

Voor wat betreft gezondheidsklachten hebben we bij volwassenen met bilaterale CP meer chronische pijnklachten, moeheid en depressieve symptomen gevonden in vergelijking met de algemene bevolking. Personen met ernstige moeheid gaven meer depressieve symptomen aan. Ook vonden we bij volwassenen met bilaterale CP een lage aërobe fitheid ten opzichte van een gezonde referentiegroep. De mate van dagelijkse lichamelijke activiteit was eveneens verminderd in deze groep, vooral bij mensen met een laag niveau van motorisch functioneren. Bij mannen was een lagere aërobe fitheid gerelateerd aan meer klachten van moeheid en bij vrouwen aan een lagere door henzelf gerapporteerde lichamelijke activiteit. Het geschatte tienjaarsrisico op sterfte door hart- en vaatziekten was laag bij volwassenen met bilaterale CP. Wel vonden we diverse biologische en leefstijlgerelateerde risicofactoren voor hart- en vaatziekten, vooral (pre)hypertensieve bloeddrukwaarden. Ook vonden we een trend wat betreft de relatie tussen lichaamsvet en het tienjaarsrisico op sterfte door hart- en vaatziekten. Dagelijkse lichamelijke activiteit en aërobe fitheid waren niet gerelateerd aan het tienjaarsrisico op sterfte door hart- en vaatziekten.

Tot zover hebben we geen duidelijke aanwijzing gevonden voor een centrale rol van dagelijkse lichamelijke activiteit in het ontwikkelen of verergeren van klachten conform de cyclus van deconditioneren volgens Durstine. Echter op basis van dit en ander onderzoek op dezelfde dataset lijkt aërobe fitheid wel een rol te spelen in deze cyclus en zou daarmee een mogelijk aangrijpingspunt kunnen zijn voor behandeling van mensen met bilaterale CP.

Het onderzoek naar volwassenen met unilaterale CP richtte zich voor wat betreft gezondheidsklachten vooral op dagelijkse lichamelijke activiteit. In deze minder ernstig aangedane groep dan bilaterale CP vonden we een vergelijkbaar niveau van dagelijkse lichamelijke activiteit als bij leeftijdgenoten zonder lichamelijke beperking.

Op het gebied van participatie ervaren volwassenen met bilaterale CP in verschillende domeinen moeite, vooral bij mobiliteit, huishouden en vrije tijd. Chronische pijn en moeheid lijken niet geassocieerd te zijn met dagelijkse activiteiten en sociale participatie bij volwassenen met bilaterale CP. Daarentegen was de participatie van volwassenen met unilaterale CP vergelijkbaar met die van leeftijdgenoten zonder lichamelijke beperking, behalve dat volwassenen met unilaterale CP minder vrijwilligerswerk en meer niet intensieve vrijetijdsactiviteiten (bijvoorbeeld lezen en tv-kijken) verrichtten.

Ondanks hun relatief jonge leeftijd (maximaal 45 jaar), hebben alle volwassenen met bilaterale CP in dit onderzoek één of meer gezondheidsklachten of participatieproblemen. Vergeleken met de algemene bevolking ervaren zij een lage gezondheidsgerelateerde kwaliteit van leven voor lichamelijke functies, maar niet voor psychische functies.

Self-efficacy (geloof in eigen kunnen) was vergelijkbaar tussen volwassenen met bilaterale CP en een referentiegroep. Op de subschaal ‘doorzetten bij tegenslag’ scoorden de volwassenen met bilaterale CP echter hoger. Self-efficacy was gerelateerd aan betere sociale participatie en de subschaal ‘inspannen om een doel te bereiken’ aan gezondheidsgerelateerde kwaliteit van leven bij volwassenen met bilaterale CP.

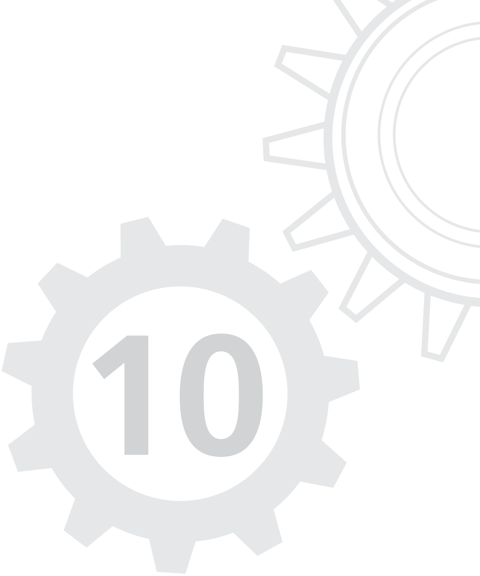
Analyses van subgroepen laten zien dat volwassenen met bilaterale CP en een lager niveau van motorisch functioneren vaker depressieve symptomen hebben, meer moeite met dagelijkse activiteiten en sociale participatie hebben en een lagere lichamelijke gezondheidsgerelateerde kwaliteit van leven aangeven. Vrouwen met bilaterale CP hebben zowel een lagere aërobe fitheid als een lager tienjaarsrisico op sterfte door hart- en vaatziekten dan mannen met bilaterale CP. Voor andere gezondheidsklachten en participatiedomeinen hebben we geen verschillen gevonden tussen mannen en vrouwen in beide onderzoeksgroepen. Ook zijn leeftijd en opleidingsniveau in dit onderzoek niet gerelateerd aan gezondheidsklachten en participatie bij volwassenen met unilaterale of bilaterale CP.

Discussie

In hoofdstuk 8 bediscussiëren we de onderzoeksresultaten mede in het licht van eerder onderzoek. Methodologische aspecten van het onderzoek worden eveneens besproken, zoals de onderzoeksopzet en generaliseerbaarheid van de resultaten.

Vervolgonderzoek zou zich kunnen richten op het verder ontrafelen van onderliggende mechanismen van gezondheidsklachten, zoals pijn, om aangrijpingspunten te vinden voor preventie en behandeling. Ook interventiestudies zijn noodzakelijk om de effectiviteit van nieuwe behandelingen bij volwassenen met CP vast te stellen (zie hoofdstuk 8 voor een beschrijving van verschillende opties).

Tot slot, CP is een levenslange aandoening en het frequent en gelijktijdig voorkomen van gezondheidsklachten en participatieproblemen vergen een holistische - en levensloopbenadering in de revalidatiezorg. De resultaten van dit proefschrift geven een aanzet tot het ontwikkelen van preventieve maatregelen en interventieprogramma's. Bijvoorbeeld training van vaardigheden ter verbetering van self-efficacy, met als uiteindelijk doel betere participatie en kwaliteit van leven. Training van aërobe fitheid zou de spiraal van deconditioneren kunnen doorbreken en zowel gezondheid als participatie ten goede kunnen komen. Gezien het gelijktijdig voorkomen van gezondheidsproblemen is behandeling van symptoomclusters relevant. Op basis van voorgaande studies worden behandelmogelijkheden besproken, onder andere cognitieve gedragsmatige therapie. Verder is educatie voor medici en paramedici uit de eerste en tweede lijn van belang, bijvoorbeeld om kennis over het diagnosticeren van gezondheidsklachten te verbeteren, zodat men tijdig kan ingrijpen. Met een dergelijke aanpak hopen wij een bijdrage te leveren aan een levensloopbenadering voor mensen met CP. Wij pleiten voor een overstijgende benadering waarmee langetermijnklachten worden voorkomen en een optimale participatie wordt bereikt.



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Dankwoord (Acknowledgements)
Curriculum vitae
List of publications
PhD portfolio

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CURRICULUM VITAE

Wilma van der Slot was born in The Netherlands. She passed her athenaeum at College Leeuwenhorst in Noordwijkerhout in 1988. The same year she attended Leiden University to study biology and finished the propaedeutic exam in 1989. Subsequently, she began to study medicine at Leiden University and performed her master's research projects at the Department of Neurophysiology, Leiden University and at the Department of Pharmacology, Exeter College, Oxford University, England. In 1994 she received her doctoranda degree (MSc) in medicine and began her medical internships.

In 1997 she obtained her Doctor of Medicine degree from Leiden University and started as both a resident in Medical Microbiology and a PhD researcher in Clinical Virology at the Department of Medical Microbiology, Leiden University Medical Centre. In 1998 she switched to Physical and Rehabilitation Medicine and worked at The Foundation of Rehabilitation Medicine Zeeland (currently Revant) in Goes and Terneuzen. In 1999 she became a resident in Physical and Rehabilitation Medicine at Rijnlands Rehabilitation Centre, Katwijk and Sophia Rehabilitation, The Hague and Gouda. She obtained the degree of Medical Doctor in Physical and Rehabilitation Medicine in 2003. Concurrently, she became certified by the European Board of Physical and Rehabilitation Medicine.

In 2003 she began working as both a clinician (her principal role) and a PhD researcher at Rijndam Rehabilitation Centre and the Department of Rehabilitation Medicine and Physical Therapy, Erasmus MC, University Medical Centre in Rotterdam. Her PhD research 'Health issues and participation in adults with cerebral palsy', is part of the Department's larger research programmes 'Transition and Lifespan Research' and 'MoveFit'. From 2003 to 2006 she worked as a staff member at the adult inpatient clinic of neurorehabilitation of Rijndam Rehabilitation Centre. Subsequently, she started at the outpatient clinic of neurorehabilitation. Amongst other things, she is involved in the treatment of adults with cerebral palsy and the young adult team for specialised rehabilitation care during the transition into adulthood. From 2006 onwards she helped implement the young adult team and participated in the Centre's development group 'Cerebral Palsy and Young Adult Team'.

Furthermore, she participates in TransitieNet and was an active member of the committees 'Transition in Action' and 'TransitieNet Moves On' aimed at sharing expertise, development and implementation of transition and lifespan care for young people with a chronic disability in The Netherlands. She is a member of the VRA, ISPRM, Patient Association BOSK, and the American Academy for Cerebral Palsy and Developmental Medicine and its Life Span Care committee.

LIST OF PUBLICATIONS

Van der Slot WMA, Roebroeck ME, Nieuwenhuijsen C, Bergen MP, Stam HJ, Burdorf A, van den Berg-Emons HJG, the MoveFit and Lifespan Research Group. Cardiovascular disease risk in adults with spastic bilateral cerebral palsy. J Rehabil Med. 2013 Jul 4. doi: 10.2340/16501977-1185. [Epub ahaed of print]

Slaman J, Bussmann J, van der Slot WMA, Stam HJ, Roebroeck ME, van den Berg-Emons HJG, the Transition and Lifespan Research Group South West Netherlands. Physical strain of walking relates to activity level in adults with cerebral palsy. *Submitted for publication.*

Van der Slot WMA, Nieuwenhuijsen C, van den Berg-Emons HJG, Bergen MP, Hilberink SR, Stam HJ, Roebroeck ME. Chronic pain, fatigue, and depressive symptoms in adults with spastic bilateral cerebral palsy. *Developmental Medicine & Child Neurology* 2012; 54: 836-42.

Slaman J, Roebroeck ME, van Meeteren J, van der Slot WMA, Reinders-Messelink HA, Lindeman E, Stam HJ, van den Berg-Emons HJG. Learn 2 Move 16-24: effectiveness of an intervention to stimulate physical activity and improve physical fitness of adolescents and young adults with spastic cerebral palsy; a randomized controlled trial. *BMC Pediatrics* 2010; 10: 79.

Van der Slot WMA, Nieuwenhuijsen C, van den Berg-Emons HJG, Wensink-Boonstra AE, Stam HJ, Roebroeck M, the Transition Research Group South West Netherlands. Participation and health-related quality of life in adults with spastic bilateral cerebral palsy and the role of self-efficacy. *Journal of Rehabilitation Medicine* 2010; 42: 528-35.

Wiegerink DJHG, Roebroeck ME, van der Slot WMA, Stam HJ, Cohen-Kettenis PT, the Transition Research Group South West Netherlands. Importance of peers and dating in the development of romantic relationships and sexual activity of young adults with cerebral palsy. *Developmental Medicine & Child Neurology* 2010; 52: 576-82.

Nieuwenhuijsen C, van der Slot WMA, Dallmeijer AJ, Janssens PJ, Stam HJ, Roebroeck ME, van den Berg-Emons HJG, the Transition Research Group South West Netherlands. Physical fitness, everyday physical activity, and fatigue in ambulatory adults with bilateral spastic cerebral palsy. *Scandinavian Journal of Medicine & Science in Sports* 2010; 21: 535-42.

Buffart LM, van den Berg-Emons HJG, van Mechelen W, van Meeteren J, van der Slot WMA, Stam HJ, Roebroeck ME. Promoting physical activity in an adolescents and a young adult with physical disabilities. *Disability and Health Journal* 2010; 3: 86-92.

Nieuwenhuijsen C, van der Slot WMA, Beelen A, Arendzen JH, Roebroeck ME, Stam HJ, van den Berg-Emons HJG, the Transition Research Group South West Netherlands. Inactive lifestyle in adults with bilateral spastic cerebral palsy. *Journal of Rehabilitation Medicine* 2009; 41: 375-81.

Van der Slot WMA, Roebroeck ME, Landkroon AP, Terburg M, van den Berg-Emons HJG, Stam HJ. Everyday physical activity and community participation of adults with hemiplegic cerebral palsy. *Disability and Rehabilitation* 2007; 29: 179-89.

Risch A, Smelt V, Lane D, Stanley L, van der Slot WMA, Ward A, Sim E. Arylamine N-acetyltransferase in erythrocytes of cystic fibrosis patients. Pharmacology Toxicology 1996; 78:235-40.

PHD PORTFOLIO

Erasmus MC
Universitair Medisch Centrum Rotterdam

Summary of PhD training and teaching activities

Name PhD student: W.M.A. van der Slot

Erasmus MC Department: Rehabilitation Medicine & Physical Therapy

PhD period: 2003–2012 (part-time)

Promotor: Prof. dr. H.J. Stam

Supervisors: Dr. M.E. Roebroeck, Dr. H.J.G. van den Berg-Emons

	Year	**Workload (ECTS)**
1. PhD training		
General academic skills courses		
• Presenteren en informatieoverdracht; Postgrade, Utrecht	2009	0.4
• How to write and read a medical paper; The Erasmus Summer Programme, Erasmus MC, Rotterdam	2006	0.5
Research skills courses		
• Logistische regressie en analyse van overlevingsduren; Afdeling Epidemiologie & Biostatistiek (EpidM), VU medisch centrum, Amsterdam	2010	1.9
• Statistische begrippen in de medische literatuur deel 1 en 2; Pfizer Nascholing en Praktijkondersteuning, Leiden	2008	0.3
In-depth courses		
• Cerebrale parese, bewegen en sport; Werkgroep Bewegen & Sport van de VRA, Emmen	2011	0.2

	Year	Workload (ECTS)
• Scholingsprogramma revalidatiegeneeskunde: Cerebrale parese; VRA & PAOG-Heyendael, Amsterdam	2011	0.4
• Scholingsprogramma revalidatiegeneeskunde: Revalidatie bij gewrichtsziekten; VRA & PAOG-Heyendael, Amsterdam	2010	0.2
• Pijn: nieuwe inzichten in de ontwikkeling en behandeling van acute en chronische pijn; Boerhaave Commissie Postacademisch Onderwijs; Leiden	2009	0.2
• 3rd Dutch - UK Rehabilitation Meeting: 'Ageing'; VRA & BSRM, Ermelo	2008	0.3
• Sport- en beweegprogramma's: Effectiviteit in de klinische praktijk; Bohn Stafleu van Loghum Cursussen & Congressen, Rotterdam	2008	0.2
• Neurologie en psychiatrie bij de oudere mens; Stichting Postuniversitair Onderwijs, Amstelveen	2008	0.2
• Scholingsprogramma revalidatiegeneeskunde: Cerebrale parese; VRA & PAOG-Heyendael, Nijmegen	2008	0.4
• Volwassenen met een cerebrale parese; ITON, Haarlem	2007	2.0
• Spasticiteit bij kinderen en volwassenen met een verstandelijke beperking: hoe te beoordelen en wat er aan te doen?; Erasmus MC, Rotterdam	2006	0.2
• Klinische neuropsychologie volwassenen; ITON, Haarlem	2004	3.9
• Toegepaste neuroanatomie en neurofysiologie; ITON, Haarlem	2003	0.3
Presentations		
• Mini-symposium 'It is my life. Innovation of care for young adults with childhood onset disabilities'. Presentation 'Goals and methods of a young adult team'; VRA Annual Congress 2012, Noordwijkerhout	2012	0.3
• Presentatie 'Moeheid, pijn en depressieve klachten bij volwassenen met cerebrale parese'; Jubileum Congres 60 jaar BOSK, Arnhem	2012	0.4
• Free paper presentation 'Pain in adults with spastic bilateral cerebral palsy'; 66th Annual Meeting of the AACPDM, Toronto, ON, Canada	2012	0.6
• Referaat 'Cardiovasculaire risicofactoren: achtergronden en onderzoek bij volwassenen met cerebrale parese'; Regionale refereeravond revalidatie, Rotterdam	2012	0.6

	Year	Workload (ECTS)
• Preconference session 'Lifespan care – a global perspective': presentation and handout 'Lifespan care in The Netherlands from a clinical perspective: transition to adulthood of young people with a childhood-onset disability and care for adults with cerebral palsy'. Free paper presentation 'Cardiovascular disease risk and relationships with waist circumference, aerobic fitness and daily physical activity in adults with spastic bilateral cerebral palsy'; 65th Annual Meeting of the AACPDM, Las Vegas, NV, USA	2011	1.6
• Presentation 'Cardiovascular disease risk in adults with spastic bilateral cerebral palsy'; Research meeting, Department of Rehabilitation Medicine and Physical Therapy, Erasmus MC, Rotterdam	2011	0.5
• Referaat 'Vermoeidheid binnen de revalidatie: algemene inleiding'; Regionale refereeravond revalidatie, Rotterdam	2011	0.6
• Referaat 'Pijn, moeheid en depressieve symptomen bij volwassenen met spastische bilaterale cerebrale parese'; ZWN refereeravond, Rotterdam	2010	0.3
• Presentatie 'Fysieke activiteit, fitheid en moeheid bij volwassenen met spastische cerebrale parese'; Interessegroep Revalidatie van de VvBN, Universitair Medisch Centrum Groningen	2010	0.7
• Free paper presentation 'High prevalence and co-occurrence of chronic pain, fatigue and depressive symptoms in adults with spastic cerebral palsy'; 64th Annual meeting of the AACPDM, Washington, DC, USA	2010	0.6
• Workshop 'Zelfmanagement in de praktijk: gemeenschappelijk beleid en visie'; Werkconferentie Op Eigen Benen Vooruit!, Nieuwegein	2010	0.6
• Presentatie 'Polikliniek Jongvolwassenen'; Informatieavond voor leerlingen en ouders over transitie naar volwassenheid, Mytylschool De Brug, Rotterdam	2009	0.2
• Workshop 'Lifespan care session for professionals' and Free paper presentation 'Participation, health-related quality of life and self-efficacy in adults with spastic bilateral cerebral palsy'; International symposium 'Global Status Quo on Cerebral Palsy, with a View to the Future', ICPS, Dutch-ACD & BOSK, Utrecht	2009	1.0
• Presentatie 'Implementatie van een polikliniek jongvolwassenen; Sectie kinderrevalidatiegeneeskunde, Utrecht	2009	0.2
• 2 free paper presentations 'Participation, health-related quality of life and self-efficacy in adults with spastic bilateral cerebral palsy' and 'Physical activity in adults with spastic cerebral palsy'; 3rd ICPC, Sydney, Australia	2009	1.0

	Year	Workload (ECTS)
• Presentatie 'Polikliniek jongvolwassenen en transitiezorg'; Informatieavond voor leerlingen en ouders over toekomstmogelijkheden, Mytylschool De Brug, Rotterdam	2008	0.2
• Workshop 'Transition from childhood to adulthood: who cares?' Presentation: 'Self-efficacy of young adults with a disability'; 3rd Dutch - UK Rehabilitation Meeting 'Ageing'; VRA & BSRM, Ermelo	2008	0.4
• Presentatie 'Transitiezorg voor jongeren en jongvolwassenen met een aangeboren of jongverworven beperking'; ambulante begeleiding en leerkrachten van VSO, Mytylschool De Brug, Rotterdam	2007	0.2
• Workshop 'Zelfstandig leven is vast ook iets voor jou!'; BOSK Landelijke Themadag 2007, Veldhoven	2007	0.3
• Instructie cursus 'Transitiezorg: Revalidatiebehandeling voor jongvolwassenen met een aangeboren of jongverworven beperking'; 3rd Dutch-ACD symposium, Utrecht	2007	0.3
• Instructional Course 'Specialised rehabilitation care for young adults with a disability'; 19th Annual Meeting of the EACD, Groningen	2007	0.4
• Presentatie in de workshop 'Transitie: hoe kan het beter? Praktijkvoorbeelden van Zorginnovaties'; Conferentie Klaar?...Over!, Kenniskring Transities in Zorg Hogeschool Rotterdam, Erasmus MC & ZONMW, Rotterdam	2007	0.6
• Poster presentation 'Everyday physical activity in young adults with hemiplegic cerebral palsy'; VRA Jaarcongres 2005, Arnhem	2005	0.6
• Workshop 'Jongeren met cerebrale parese op weg naar volwassenheid'; Landelijke dag van de werkgroep spasticiteit van de BOSK, Utrecht	2004	0.4
• Presentatie 'Studie protocol: Dagelijks functioneren en fitheid bij volwassenen met spastische cerebrale parese'; Research meeting, Department of Rehabilitation Medicine and Physical Therapy, Erasmus MC, Rotterdam	2004	0.3
• Referaat 'Dagelijkse lichamelijke activiteit en fitheid bij volwassenen met een spastische cerebrale parese. Is een interventie ter bevordering van een meer actieve leefstijl wenselijk?'; Regionale refereeravond revalidatie, Rotterdam	2003	0.3
Online contributions		
• Deelname aan Discussion Podcast of paper 'Chronic pain, fatigue, and depressive symptoms in adults with spastic bilateral cerebral palsy'; Developmental Medicine & Child Neurology podcasts	2012	0.2

	Year	Workload (ECTS)
• Website 'Cerebrale parese', Rijndam revalidatiecentrum: coördinatie van en bijdrage aan het onderdeel (jong)volwassenen met cerebrale parese	2010–2011	1.0
• Website 'Op Eigen Benen': Toolkit van het actieprogramma Op Eigen Benen Vooruit: Interventies en instrumenten gericht op de organisatie van zorg: Polikliniek Jongvolwassenen – organisatie en praktijkervaring	2010	0.2
International conferences		
• 66th Annual Meeting of the AACPDM, Toronto, ON, Canada	2012	0.8
• 65th Annual Meeting of the AACPDM, Las Vegas, NV, USA	2011	0.8
• 64th Annual meeting of the AACPDM, Washington, DC, USA	2010	0.8
• International symposium 'Global Status Quo on Cerebral Palsy, with a View to the Future' of the ICPS, Dutch-ACD & BOSK, Utrecht, The Netherlands	2009	0.2
• 3rd ICPC, Sydney, Australia	2009	1.2
• 61st Annual meeting of the AACPDM, Vancouver, BC, Canada	2007	0.9
• 19th Annual Meeting of the EACD, Groningen, The Netherlands	2007	0.9
Seminars and guideline meetings		
• Seminar 'Cerebral palsy: the role of fitness training and physical activity in rehabilitation'; VU medisch centrum, Amsterdam	2011	0.1
• Vermoeidheid in de Revalidatie - Actueel bewegingswetenschappelijk revalidatieonderzoek; Interessegroep Revalidatie VvVB, Universitair Medisch Centrum Groningen	2010	0.1
• TransitieNet Bijeenkomst 'Actieve leefstijl en Sportstimulering & Handvatten voor het opzetten van een polikliniek jongvolwassenen'; Den Haag	2008	0.1
• Richtlijnbijeenkomst 'Diagnostiek en behandeling van kinderen met spastische cerebrale parese'; Kwaliteitsinstituut voor de gezondheidszorg – CBO, Utrecht	2006	0.2
Research meetings		
• Research meetings; Department of Rehabilitation Medicine and Physical Therapy, Erasmus MC, Rotterdam	2003–2012	2.3
• ZWN-refereeravonden; Revalidatie Rotterdam/Leiden/Den Haag	2003–2012	0.4

	Year	Workload (ECTS)
Other		
• 'Lifespan care: transitie naar volwassenheid en volwassenen met cerebrale parese', Cursusboek 'Cerebrale parese; stand van zaken en nieuwe ontwikkelingen'; VRA & PAOG-Heyendael, ISBN: 978-90-373-0391-9, p.191-198	2011	0.5
• Adviseren aflevering Cerebrale parese van 'Je zal het maar hebben'; BNN	2009	0.2
• 'Beter begeleiden naar volwassenheid' Interview in: Revalidatie Magazine 15 (1): 16-17	2009	0.1
• 'Op weg naar een zelfstandig leven' Interview in: BOSK Magazine 3: 8-9	2008	0.1
• Deelname aan Expertmeeting actie- en implementatieprogramma 'Op Eigen Benen Vooruit!'; Kwaliteitsinstituut voor de gezondheidszorg - CBO, Utrecht	2008	0.3
• Bijdrage aan factsheets en modulaire behandelprogramma polikliniek jongvolwassenen Rijndam revalidatiecentrum/Erasmus MC	2006–2007	1.0
2. Teaching activities		
Supervising practicals and lecturing		
• Scholingsprogramma revalidatiegeneeskunde 'Cerebrale parese; stand van zaken en nieuwe ontwikkelingen'. Onderdeel: 'Lifespan care: transitie naar volwassenheid en volwassenen met cerebrale parese'; VRA & PAOG-Heyendael, Amsterdam	2011	0.6
• 'Cursus Volwassenen met cerebrale parese': begeleiden van practica en docent van: 'Vermoeidheid en activiteit' en 'Maatschappelijk functioneren'; ITON, Haarlem	2008	1.4
Total ECTS		**39.2**

AACPDM: American Academy for Cerebral Palsy and Developmental Medicine; BOSK: Dutch Association of Physically Disabled Persons and their Parents; BSRM: The British Society of Rehabilitation Medicine; Dutch-ACD: Dutch Academy of Childhood Disability; EACD: European Academy of Childhood Disability; ICPC: International Cerebral Palsy Conference; ICPS: International Cerebral Palsy Society; ITON: Instituut voor Toegepaste Neurowetenschappen; VvBN: Vereniging voor Bewegingswetenschappen Nederland; VRA: The Netherlands Society of Physical and Rehabilitation Medicine; ZWN: Zuid-West Nederland