Adults with Cerebral Palsy
Studies on physical activity, fitness and experienced problems

Channah Nieuwenhuijsen

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Adults with Cerebral Palsy
Studies on physical activity, fitness and experienced problems

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Studies naar dagelijkse lichamelijke activiteit, fitheid en ervaren problemen

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CONTENTS

CHAPTER 1 Introduction

CEREBRAL PALSY

Cerebral palsy (CP) is a frequently occurring condition in childhood: in the Netherlands children with CP account for approximately 50% of pediatric rehabilitation patients.[1] 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 1000 live births in Europe.[3] In a Dutch population-based study, the average prevalence over 12 birth years (1977-1988) was 1.51 per 1000 inhabitants. The CP prevalence rose significantly over time: from 0.77 per 1000 inhabitants in 1977-1979 to 2.44 per 1000 in 1986-1988.[4] Over the past decades, life expectancy for well-functioning adults with CP has increased and is close to that of the unaffected population.[5]

CEREBRAL PALSY IN ADULTHOOD

Despite the high survival rates for persons with CP, consequences of CP in adulthood are often poorly understood.[6] As described by the definition of CP, problems can occur in several areas. Furthermore, with ageing, health issues such as increased risk for cardiovascular disease and age-related decline in function are increasingly important to consider. At the same time, as persons with CP enter adulthood, attendance to rehabilitation care seriously declines, despite all the physical, social and emotional changes that may occur during this transition into adulthood.[7] Several studies report decreased contact with the health care system following completion of formal education and care is often disrupted when disabled persons enter adulthood.[8,9]

However, many persons with CP return to rehabilitation care at adult age for treatment of worsening health problems such as fatigue and also contractures and pain.[10,11] From studies in Norway in unilateral and bilateral adults with CP, we know that fatigue is a common problem in CP: 30% of them experienced substantial fatigue, and 17% reported chronic fatigue, mainly those with bilateral CP.[12-14] Adults with CP can also experience problems in other areas: their participation in daily life and their experienced health-related quality of life (HRQoL) is shown to be lower than in the general population. For adolescents and adults with CP, and particularly those with bilateral CP, restrictions in participation have been reported in several areas, such as outdoor mobility, recreation, self-care and employment.[15-17] HRQoL is also lower than in the general population, especially in the physical domain.[17-19]

LEVEL OF EVERYDAY PHYSICAL ACTIVITY AND PHYSICAL FITNESS

It could be hypothesized that low levels of everyday physical activity (PA) and low levels of physical fitness may contribute to the worsening of health problems when persons with CP grow into adulthood. In the general population, PA and physical fitness are considered of major benefit to a healthy lifestyle[20,21] and are related to amongst all cardiovascular disease and diabetes mellitus.[22-24] In persons with CP, PA and physical fitness may be even more important in order to offset the decline in function that might occur with aging and deterioration of CP-related impairments such as reduced range of motion and increased spasticity or pain.[21]

Despite the expectation that persons with disabilities are at a high risk for an inactive lifestyle[25], only limited objective information is available regarding level of everyday PA of adults with CP. There is evidence that diplegic children[26] and adolescents with certain forms of CP[27] are less physically active than able-bodied age-mates. Particularly children and adolescents with a lower level of gross motor functioning had lower levels of everyday PA.[27,28] In adults, van der Slot et al.[16] reported no differences in level of everyday PA between persons with unilateral spastic CP and able-bodied age-matched controls. Objective data regarding the level of everyday PA in adults with bilateral spastic CP are lacking.

Concerning physical fitness, again, research in CP has focused almost entirely on children and adolescents, showing lower levels of physical fitness in those with CP.[21] Research in adults suggests that physical fitness, operationalized as peak oxygen uptake, appears to be lower in both ambulatory and non-ambulatory adults with CP.[29,30] However, these studies were performed in a small sample and specific data on those with bilateral CP are not available.

Besides its possible worsening effects on health problems, level of everyday PA and physical fitness could also play an important role in participation and HRQoL. Only few studies in CP have addressed the role of PA[18,31] and no studies are available on the role of physical fitness in participation and HRQoL. In adults[18] and adolescents[31], weak associations have been reported between PA and the various domains of HRQoL, with the strongest relations found with the physical functioning domain.

Because of amongst all physical complaints, many persons with CP return to rehabilitation care at adult age. Therefore, it is important to optimize this rehabilitation care for adults with CP. Level of everyday PA and physical fitness are factors at which rehabilitation care could be aimed. Next to the importance of improving these factors as indicators for health, level of everyday PA and physical fitness could also contribute to the optimalisation of rehabilitation care offered to adults with CP in other areas. When low levels of everyday PA and fitness are present in adults with CP, and these factors are related to increased health problems and low participation and HRQoL in adults with CP, interventions aimed on improving both factors may be worthwhile to secondarily increase the well-being of adults with CP in different areas, such as health problems, participation and HRQoL.

EXPERIENCED PROBLEMS

Besides everyday PA and physical fitness, other factors may contribute to the optimization of rehabilitation care for adults with CP. In early adulthood, persons with CP may experience problems, which in turn may lead to exacerbation of the health problems at higher ages. For example, having difficulties with walking long distances as an adolescent, may lead to the performance of less daily activities, which may increase symptoms of for example fatigue. These newly experienced problems may warrant further treatment.[32] However, several studies reported a discontinuity of care when young adults with physical disabilities enter adulthood and reported diminished contact with health care after leaving school.[9,33] This lack of adequate rehabilitation care, together with changes in social role and in environmental expectations as they grow into adulthood, may result in other unmet (health) needs of young adults with physical disabilities.

There is an increased awareness about the need for lifespan care of persons with CP[34-36], and the number of initiatives is growing. In this way, we can recognize the possible difficulties encountered by young adults with CP, which may lead to larger problems at higher age. Although the number of lifespan care initiatives is growing, we have limited systematic knowledge of specific problems encountered by young adults with CP during the transition to adulthood. Some studies have been published regarding limitations of adolescents with CP in daily activities and social roles[15,37]; however, further knowledge of experienced problems of young adults themselves is of great value in determining relevant topics for interventions.

AIMS AND OUTLINE OF THE THESIS

In this thesis we intended to establish modifiable factors at which rehabilitation care in adults with CP may be aimed, in order to have starting points for optimizing rehabilitation care for this group. Level of everyday PA, physical fitness and self-experienced problems may be possible contributors to an increased level of health problems, which may cause adults with CP to return to rehabilitation care. At present, we know little about PA levels and physical fitness levels in persons with CP and about the possible role of these factors in health problems, participation and HRQoL. Furthermore, little is known about problems that young adults with CP experience themselves and which may further aggravate experienced problems in well-being at later ages. To be able to optimize the rehabilitation care of adults with CP, insight in both areas is warranted. In the first part of this thesis (chapter 2, 3 and 4) we aimed to describe several studies concerning level of everyday PA, physical fitness, participation and HRQoL in adults with bilateral spastic CP aged between 25 and 45. In the second part of this thesis (chapter 5 and 6) we investigated problems experienced as reported by young adults (aged between 18 and 39) themselves and health care utilization.

Chapter 2 of this thesis describes a cross-sectional study, which we performed to quantify the level of everyday PA in both ambulatory and wheelchair-dependent adults with CP. With an accelerometry-based Activity Monitor the level of everyday PA in a 24-hour period was determined. Also, we explored relations with personal and CP-related characteristics to determine whether subgroups of adults with CP are particularly at risk for an inactive lifestyle.

In **chapter 3** we assessed physical fitness using progressive maximal aerobic cycle tests and we studied interrelations between physical fitness, level of everyday PA and fatigue. Peak oxygen uptake, objective and self-reported levels of PA and level of fatigue were determined in ambulatory adults with bilateral spastic CP.

Furthermore, we explored physical fitness and level of everyday PA as potential correlates of participation and HRQoL (**chapter 4**). Studying the same sample as described in chapter 3, in this chapter we wanted to determine whether levels of physical fitness and level of everyday PA are related to HRQoL and participation.

The problems experienced by young adults with CP (18-22 years) are described in a broad sense by using semi-structured interviews in **chapter 5**. In this study, we used the International Classification of Functioning to categorize the experienced problems.

In **chapter 6** we determined the unmet needs and health care utilization of (young) adults with CP (aged 18-39). The participants filled in several questionnaires to gain insight in both factors.

Finally, **chapter 7** of the thesis describes the main findings of the thesis, discusses the strengths and limitations of the studies mentioned above, followed by the clinical implications of our studies and suggestions for future research.

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CHAPTER 2

Inactive lifestyle in adults with bilateral spastic cerebral palsy

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ABSTRACT

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

Methods Fifty-six adults with bilateral spastic cerebral palsy (mean age 36.4 (standard deviation (SD) 5.8) years, 62% male) 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 characterized by: (*i*) duration of dynamic activities (composite measure, percentage of 24 h); (*ii*) intensity of activity (motility, in gravitational acceleration (g)); and (*iii*) 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; $p \leq .01$).

Conclusions Adults with bilateral spastic 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 1000 live births in Europe.[1] The prevalence in the Netherlands is 1.51 per 1000 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 characterized 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 disabled 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. (2007) 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 bilateral spastic CP. The aims of this study were, therefore: (*i*) to quantify the level of everyday PA for adults aged 25-45 years with bilateral spastic CP, and compare them with the levels of able-bodied age-mates; and (*ii*) 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 bilateral spastic CP (diplegia or quadriplegia) and age between 25 and 45 years. Exclusion criteria were full dependence on electric wheelchair propulsion, comorbidities impacting PA, contraindications to progressive maximal cycle 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 Centre 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: *(i)* low, including prevocational practical education or less; (*ii*) medium, including prevocational theoretical education and upper secondary vocational education; and (*iii*) 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, 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 × 9 × 4.5 cm; weight 700 g) 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 × 1.5 × 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: (*i*) 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); (*ii*) number of transitions (includes all transitions except lying transitions between prone and supine positions); (*iii*) intensity of activities: (*iiia*) mean motility (in gravitational acceleration (g)), which reflected both duration and intensity of activity; (*iiib*) motility during walking; (*iiic*) motility during wheelchair propulsion; and (*iv*) 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").

Data 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= .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 (Rs). 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 ≤ .05 was considered significant.

RESULTS

Personal and cerebral palsy-related characteristics

Of 226 eligible 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≤ .01).

The mean participant age was 36.4 (standard deviation (SD) 5.8) years; 62% were male. Affected limb was evenly distributed between quadriplegics and diplegics (Table 1). Most participants (73%) had high gross motor functioning (GMFCS level I or II). Seven participants used wheelchairs; 3 GMFCS level IV participants used wheelchairs as their primary mode of ambulation, and 4 GMFCS level II participants 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 2). With regard to intensity of activities, we found a mean motility of 0.020 (SD 0.007) g.

Table 1. Personal and cerebral palsy-related characteristics.

	Participants (*n*=56)	
Age, years, mean (standard deviation)	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* (%)		
Male	35	(62)
Female	21	(38)
Limb distribution, *n* (%)		
Diplegia	30	(54)
Quadriplegia	26	(46)
GMFCS, *n* (%)*		
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* (%)#		
2 muscle groups	5	(10)
3 muscle groups	19	(38)
4 muscle groups	26	(52)
Educational level, *n* (%)		
Low	15	(27)
Medium	24	(43)
High	17	(30)
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)

* Wheelchair-users were distributed over GMFCS level II (*n*=4) and GMFCS level IV (*n*=3)

\# Spasticity was not assessed in 6 participants (*n*=50)

GMFCS= Gross Motor Functioning Classification System

Motility during walking was 0.155 (SD 0.037) g, and motility during wheelchair propulsion was 0.034 (SD 0.011) g.

Table 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).

Table 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)	**GMFCS I (*n*=13)**	**GMFCS II (*n*=28)**	**GMFCS III-IV (*n*=15)**
Duration of static activity (% of 24 h)	91.9 (3.7)	89.7 (2.6)[a]	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)	**8.1 (3.7)**	**10.3 (2.6)**[a]	**8.3 (3.7)**[c]	**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*	**0.020 (0.007)**	**0.024 (0.006)**[a]	**0.020 (0.007)**[c]	**0.015 (0.005)**
Motility during walking, g*	0.155 (0.037)	0.168 (0.030)	0.160 (0.035)	0.133 (0.042)
Motility during wheelchair propulsion, g*	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	**16 (11)**	**21 (7)**	**17 (13)**	**12 (9)**
Periods of greater than 5 min continuous dynamic activities	**1 (1)**	**2 (2)**[b]	**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)

* 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).

[a] Significant difference between GMFCS level I and GMFCS level III-IV at p≤ .01.

[b] Significant difference between GMFCS level I and GMFCS level III-IV at p≤ .05.

[c] Tendency for difference between GMFCS level II and GMFCS level III-IV at p< .10.

Transitions: all transitions between postures except between lying transitions. Associations with personal and CP-related characteristics were explored for bolded variables. GMFCS= Gross Motor Functioning Classification System. VAS=Visual Analogue Scale.

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 2). Level of satisfaction was not related to the 3 main aspects of level of everyday PA (Rs = –0.13 to 0.09).

Table 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

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≤ .01), and significantly lower mean motility (0.020g vs 0.027g; p≤ .01) (Figure 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≤ .01). For men, this difference was not statistically significant (7.8% vs 9.4%, p= .11). In both women and men, mean motility was lower compared with able-bodied age-mates (women: 0.021g vs 0.028g, p= .03; men: 0.019g vs 0.027g, p≤ .01).

Factors associated with level of everyday physical activity

Gross motor functioning was the only factor significantly associated with the three main aspects of level of everyday PA (Table 4). ANOVA and *post-hoc* analyses revealed significant differences between GMFCS level I and GMFCS level III/IV participants in duration of dynamic activities (p≤ .01), mean motility (p≤ .01), and number of periods of continuous dynamic activities greater than 5 min (p≤ .05). The difference in number of 1-5 min periods of continuous dynamic activities was not significant (p= .10) (Table 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= .06 and p= .09, respectively). The number of 1 to 5 minute and greater than 5 minute periods of continuous dynamic activities did not differ between GMFCS level II and GMFCS level III/IV participants (p= .32 and p= .58). In addition, differences between GMFCS level I and GMFCS II were not significant for the three main aspects of level of everyday PA.

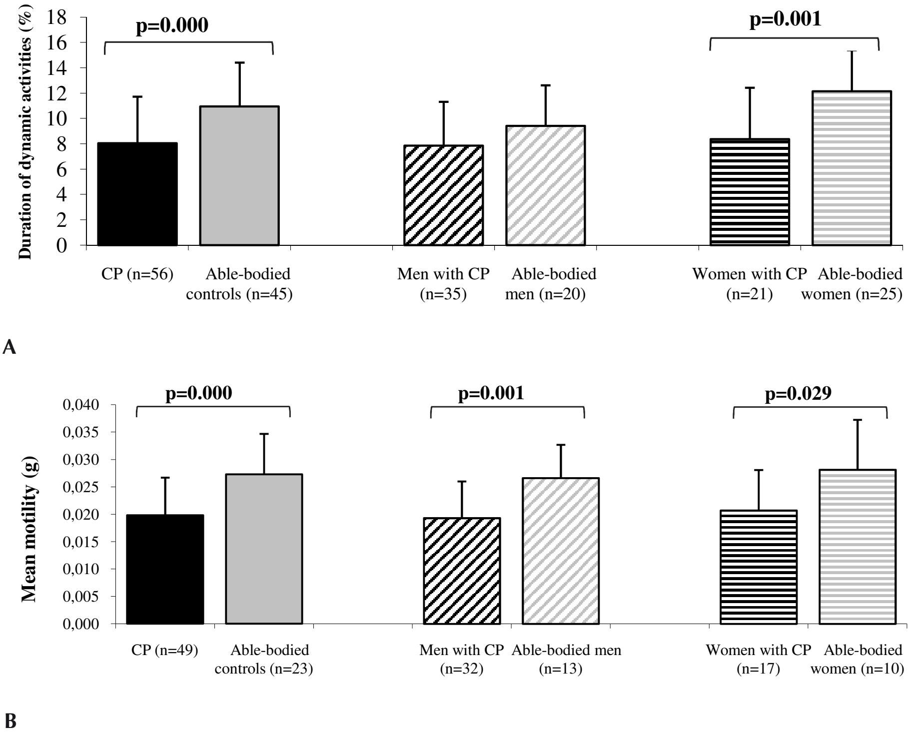

Figure 1. (a) Duration of dynamic activities in adults with bilateral spastic cerebral palsy and able-bodied age-mates, as percentage of a 24-h period. **(b)** Intensity of activities (mean motility) in adults with bilateral spastic cerebral palsy and in able-bodied age-mates, expressed in gravitational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

Table 4. Spearman correlations (Rs) 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≤ .01.

GMFCS= Gross Motor Functioning Classification System. A higher GMFCS level indicates a lower level of gross motor functioning.

DISCUSSION

This is the first study objectively measuring level of everyday PA in adults with bilateral spastic CP. We have demonstrated that adults with bilateral spastic 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 unilateral spastic 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 unilateral spastic 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. (2007) among adults with unilateral spastic 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 no to have an effect on study results. Fourth, we may have overestimated the level of everyday PA because of selection bias. Adults with CP who are interested in PA and fitness (and may therefore have had a higher level of everyday PA) may have been more likely to participate than those with less interest and lower PA.

Implications for treatment

Our study shows that adults with bilateral spastic 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 aging 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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CHAPTER 3

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

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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 bilateral spastic CP (mean age 36.4 ± 5.8 years; 69% male; 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 (Rp= 0.61, p= .03) and in men to higher levels of fatigue (Rp= -0.37, p= .05). Other relationships were not significant.

Conclusions 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 aging and deterioration of CP-related impairments such as reduced range of motion and increased spasticity or pain.[5] For example, Opheim et al. (2009)[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 hypothesized 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. (1990)[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. (1998)[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 bilateral spastic 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 bilateral spastic 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 centers 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 bilateral spastic 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 bilateral spastic CP. All participants gave written informed consent to participate. This study was approved by the Medical Ethics Committee of the Erasmus Medical Centre 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 ≥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 min 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 s 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. (2002)[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]

Data 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 ± 3.7% vs 8.5 ± 3.7%, p= .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 (Rp) 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 ≤ .05 to indicate significance.

RESULTS

Study sample

Forty-two adults with bilateral spastic CP with a mean age of 36.4 ± 5.8 years participated (Table 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%).

Physical fitness

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

Table 2 describes physical fitness for the total group and for gender subgroups. Average aerobic capacity, defined as VO_{2peak}, was 2.40 ± 0.39 L/min for men and 1.73 ± 0.32 L/min for women, which was significantly lower than age-based and gender-based reference values for sedentary Dutch men (3.18 ± 0.25 L/min, p= .03)

Table 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* (%)*						
2 muscle groups	4	(10)	4	(14)	0	(0)
3 muscle groups	15	(37)	11	(39)	4	(31)
4 muscle groups	22	(53)	13	(17)	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)

* Spasticity was not assessed in one male participant, *n*=41 (total); *n*=28 (men).

GMFCS, Gross Motor Functioning Classification System.

and women (2.15 ± 0.30 L/min, p< .01). Men had higher levels of physical fitness than women, also after correction for body mass (p< .01).

Level of everyday physical activity and fatigue

On average, objective PA was 8.6 ± 3.5%, which resembles 124 min of PA per day (Table 2). The participants were significantly less active than able-bodied age mates (10.9 ± 3.5%, p< .01). Self-reported PA was 14.8 ± 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 ≤ FSS score <5.1).

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

Figure 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: Rp= 0.26, p= .18; women: Rp= 0.26, p= .38). Women with CP who had higher levels of self-reported PA had higher levels of physical fitness (Rp= 0.61, p= .03) (Figure 1(b)).

Table 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	.000[†]
% of reference values[‡]	77±13	76±13	81±12	
VO_{2peak} (mL/kg/min)	31.5±6.3	33.2±6.2	27.9±4.8	.010[†]
% of reference values[‡]	88±17	87±19	90±15	
Peak power output (W)	144±30	153±26	124±30	.003[†]
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[§]	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	.543
% of reference values[#]	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	.754
Fatigue				
FSS score	4.1±1.3	4.1±1.2	4.0±1.5	.739
Not fatigued (FSS score <4.0)	50%	48%	54%	
Fatigued (4.0 ≤ 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 presented.

[†] Indicates a significant difference at p≤ .05.

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

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

[#] 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.

With regard to fatigue, we found that men with higher levels of physical fitness reported less fatigue (Rp= -0.37, p= .05) (Figure 2). In women, no relationship was found. Relationships between objective PA and fatigue were not significant in both men (Rp= -0.16, p= .42) and women (Rp= 0.29, p= .34). Also, self-reported PA was not associated with fatigue (Rp= -0.15, p= .44 and Rp= -0.13, p= .66, respectively). Additional analyses with VO_{2peak} corrected for body mass (mL/kg/min) showed no significant relationships among physical fitness, objective PA, self-reported PA and fatigue.

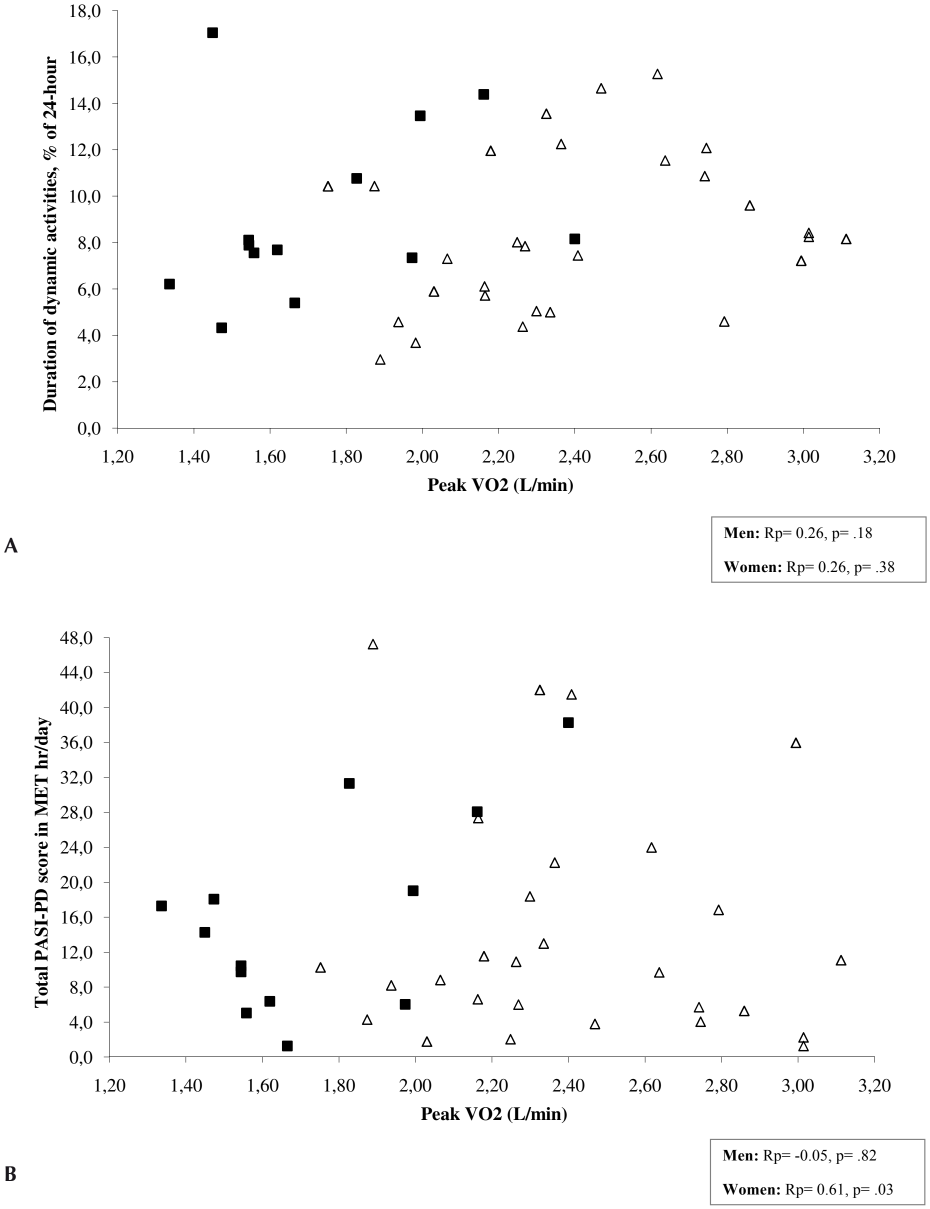

Figure 1. (a) Relationship between physical fitness and the objective level of physical activity. **(b)** Self-reported level of physical activity for men (△) and women (■).
Rp, Pearson correlation coefficient; PASIPD, Physical Activity Scale for Individuals with Physical Disabilities; MET, metabolic equivalent.

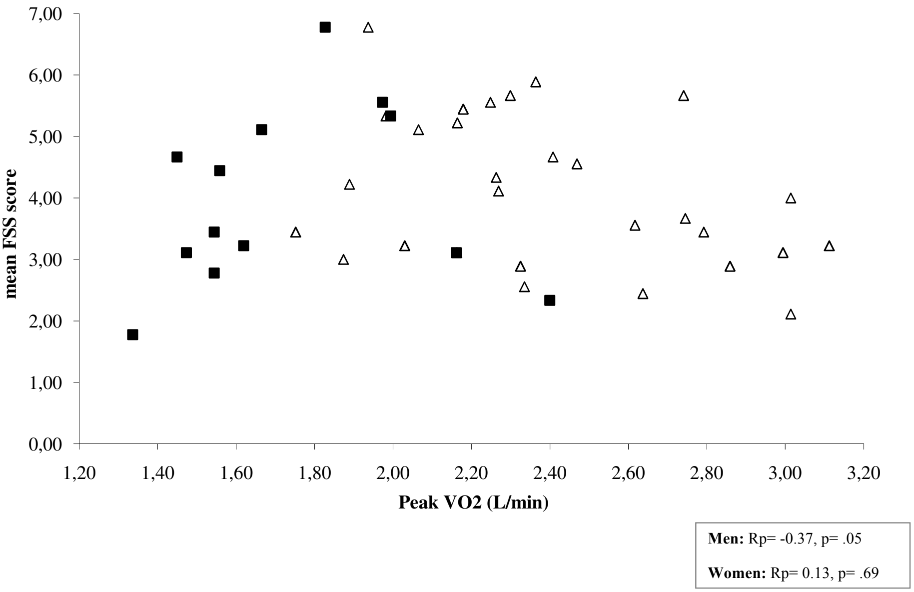

Figure 2. Relationship between physical fitness and fatigue for men (△) and women (■).
FSS, Fatigue Severity Scale; Rp, Pearson correlation coefficient.

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 localized 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 favorable effects on physical fitness in both adults[35] and children with CP.[30,32]

Relationships among 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, heart rate 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. (2009)[9] hypothesized 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 hypothesize 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 behavioral 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 among physical fitness, level of everyday PA, and fatigue. The apparent lack of consistent relationships among 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 generalized 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 PA (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 & Reibold (1990)[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 among 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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CHAPTER 4

The role of physical fitness and activity in participation and health-related quality of life in adults with cerebral palsy

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ABSTRACT

Objective To explore physical fitness and everyday physical activity (PA) as potential correlates of participation and health-related quality of life (HRQoL) in adults with bilateral spastic cerebral palsy (CP).

Methods Participation in daily activities and social roles was assessed with the Life-Habits questionnaire version 3.0 in 42 ambulatory adults with bilateral spastic CP. Physical and mental HRQoL was assessed with the SF-36. Physical fitness and level of everyday PA were measured with a progressive maximal aerobic cycle ergometer test and an accelerometry-based activity monitor, respectively. Logistic regression analyses were performed to study relationships.

Results Fifty-seven percent of participants experienced difficulties in daily activities and 36% in social roles. Vitality, physical functioning and role limitation scored lowest in physical HRQoL. Adjusted for gender and gross motor functioning, adults with higher physical fitness tended to experience less difficulties in daily activities (OR= 9.3, p= .09) and in social roles (OR= 34.1, p= .03) and had a higher physical HRQoL (OR= 24.9, p= .02). Level of everyday PA was not related to either participation or HRQoL.

Conclusions Ambulatory adults with CP often experience difficulties in participation and have low HRQoL in physical functioning. Physical fitness is related positively to these outcomes.

INTRODUCTION

Cerebral palsy (CP) is considered a childhood-onset condition. However, consequences of CP may evolve at adult age.[1] Due to factors such as medical advances, life expectancy for people with CP has increased.[2] The life expectancy for well-functioning adults with CP is close to that of the general population.[3] Therefore, insight into health issues faced by adults with CP along with ways to improve health throughout life has become increasingly important.[1,4]

In previous studies we reported a low level of everyday physical activity (PA) and a low level of physical fitness in adults with CP[5,6], indicating an inactive and possibly unhealthy lifestyle.[5-8] These aspects may be associated with restrictions in participation and low health-related quality of life (HRQoL).[9,10] Whereas objective functioning can be assessed by participation in daily activities and social roles, the concept of HRQoL is more subjectively oriented.[11,12]

Participation is defined by the World Health Organization (WHO) as “the involvement in life situations and is about what a person does in real life”. Participation is therefore strongly influenced by environment.[11] For adolescents and adults with CP, and particularly those with bilateral CP, restrictions in participation have been reported in several areas, such as outdoor mobility, recreation, self-care and employment.[13,14] Adults with unilateral CP, on the other hand have levels of community participation comparable to the general population.[8]

HRQoL refers to an individual’s perception of various aspects of his or her life that are affected by a medical condition.[11] Studies in adults[7,9,14] and adolescents[15,16] with CP have shown that HRQoL is lower in CP than in the general population, especially in the physical domain.

Factors such as level of gross motor functioning, level of education, self efficacy and fatigue have been shown to be associated with participation[4,13,14] and HRQoL.[7,14] However, lifestyle related factors such as physical fitness and PA may also be important factors. Moreover, these factors can be modified with specific interventions. Few previous studies have addressed the role of PA in participation and HRQoL in persons with CP and to our knowledge no previous studies have investigated the role of physical fitness. Gaskin and co-workers reported weak associations between self-reported PA and the various domains of HRQoL in adults with CP, with the strongest relationships found in the physical functioning domain.[9] Another study[15] has shown that walking activity is not associated with self-reported quality of life in adolescents (aged 10 to 13 years) with CP. The aim of the present study was to further explore physical fitness and level of everyday PA as potential correlates of participation and HRQoL in ambulatory adults with bilateral spastic CP.

METHODS

Study sample

Participants included patients from 10 rehabilitation centers and rehabilitation departments at university hospitals in the western and central region of the Netherlands and from the Association of Physically Disabled Persons and their Parents (BOSK). Inclusion criteria were a diagnosis of bilateral spastic CP (diplegia or quadriplegia), ambulatory with or without walking aids in daily life (Gross Motor Functioning Classification System (GMFCS) level I-III), and between 25 and 45 years old. Exclusion criteria were comorbidities impacting on PA, cardiovascular contraindications to maximal ergometer testing (such as severe heart disease), legal inability to participate in the study, inadequate comprehension of the Dutch language, and cognitive impairment preventing understanding of the study protocol.

The present study is part of a larger study[5] focusing on the level of everyday PA in 56 adults with CP. From the original cohort, 4 participants were non-ambulatory, leaving 52 eligible participants for the study. In total, 10 participants had missing values for aerobic capacity, either because of logistical problems, technical problems, impaired balance, or knee problems. Therefore, the final sample consisted of 42 adults with bilateral spastic CP. All participants gave written informed consent to participate. This study was approved by the Medical Ethics Committee of the Erasmus Medical Centre and by the medical ethical committees of all participating rehabilitation centres and hospitals.

Personal and clinical characteristics

Age, gender, limb distribution (diplegia or quadriplegia), level of gross motor functioning and level of education were determined for each participant. Level of gross motor functioning was assessed with the GMFCS.[17] The GMFCS is a five-level classification system based on spontaneous movements related to sitting and walking. Reliability and validity of the GMFCS have been established in both children[17] and adults[18] with CP. Level of education was subdivided into (1) low (prevocational practical education or less); (2) medium (prevocational theoretical education and upper secondary vocational education) or (3) high (secondary non-vocational education, higher education and university).

Participation

The short version of the Assessment of Life Habits (Life-H 3.0) was used to evaluate participation in daily activities and social roles.[19] The questionnaire consists of 69 life habits, divided in 12 categories: nutrition, fitness, personal care, communication, housing, mobility, responsibilities, interpersonal relationships, community life, education, employment and recreation. The first six categories refer to daily activities and the latter six evaluate social roles. A performance score is calculated based on the level of difficulty when performing a life habit (no difficulty, with difficulty, with substitution, or not accomplished) and the type of assistance required to perform the life habit (no help, technical assistance or adaptation, human assistance). The performance scores range from 0 to 9, in which 0 denotes total handicap (life habit is not accomplished or achieved) and 9 denotes optimal participation (life habit is accomplished without difficulty). Performance scores were generated for all domains and for the sub domains of daily activities and social roles. A mean score <8.0 signified difficulty in participation.[13] The Life-H has been shown to have moderate to good psychometric properties in a variety of groups of adults with impairments.[20,21]

Health-Related Quality of Life

We used the Medical Outcomes Study 36-item Short-form Health Survey (SF-36) to assess HRQoL.[22] The SF-36 measures HRQoL in eight domains: 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. Raw scores were converted to normalized scores ranging from 0 to 100. In addition, two main scores were calculated: a physical component summary score (PCS) and a mental component summary score (MCS), with an expected mean (± standard deviation (SD)) of 50 (± 10) obtained from the general population in the United States.[22,23] Higher scores indicate higher levels of HRQoL. The Dutch language version of the SF-36 has shown to possess good reliability and validity.[24]

Physical fitness and everyday physical activity

Aerobic fitness was measured with 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.[25] After a 3-minute warm-up period at low resistance and a 5-minute rest period, the resistance was increased every two minutes at a variable rate according to the cycling abilities of the participant. The test was terminated when the participant voluntarily stopped due to exhaustion or was unable to maintain a pedal rate of >40 rpm. More detailed descriptions of the test can be found elsewhere.[6] Aerobic capacity, defined as the mean level of oxygen uptake during the last 30 seconds of exercise (VO_{2peak}, expressed in L/min) was used as a measure of physical fitness.

The level of everyday PA was determined objectively during two consecutive weekdays (48 hours) with an accelerometry-based Activity Monitor (AM).[5,26] The AM can differentiate between stationary activities (lying, sitting and standing) and dynamic activities (walking (including running), cycling, wheelchair propulsion and general non-cyclic movement). The AM was fitted in the participant's home and participants were instructed to perform their ordinary activities with the exception of swimming or bathing. More detailed descriptions of the measurement procedures are provided elsewhere.[5,26] The level of everyday PA was calculated as the mean duration of dynamic activities measured over two days, and was expressed as percentage of 24 hours.

Data analysis

We reported participation and HRQoL in different categories or domains as mean (SD) and also the percentage of participants perceiving difficulties in daily activities and social roles (score <8.0). Since data on participation and HRQoL were not normally distributed, we dichotomized these outcome measures. For the subdomains of daily activities and social roles of the Life-H, participants were classified as either perceiving difficulties (0) or no difficulties (1). For the PCS and MCS of the SF-36, scores <50.0 were classified as a low level of HRQoL (0) and scores ≥50.0 were classified as a high level of HRQoL (1), based on the mean of the reference population in the United States.[23]

RESULTS

Forty-two ambulatory adults with CP with a mean age of 36.4 (5.8) years participated in the study (Table 1). The majority of participants were able to ambulate without assistive mobility devices (GMFCS level I or II), while others used crutches or a walker for ambulation. Mean aerobic capacity was 2.19 (0.48) L/min. Participants were dynamically active for an average of 124 min/day or 8.6 (3.5)% of a 24 hour period (Table 1).

Table 1. Personal characteristics, physical fitness and level of everyday physical activity.

	Participants (*n*=42)
Personal characteristics	
Age (*mean (SD)*)	36 years, 5 months (5y,10mo)
Gender (*n*)	29 men
Limb distribution (*n*)	22 diplegia, 20 quadriplegia
Gross motor functioning* (*n*)	11 level I, 23 level II, 8 level III-IV
Level of education (*n*)	12 high, 20 medium, 10 low
Physical fitness and level of everyday PA	
VO_{2peak}	2.19 (0.48) L/min
Duration of dynamic activities	8.6 (3.5) %

* Gross motor functioning is categorized according to the levels of the Gross Motor Functioning Classification System (GMFCS).

Table 2. Participation in daily activities and social roles in ambulatory adults with cerebral palsy.

	Mean(SD) (range 0-9)	**Perceived difficulties (%) (mean score <8.0)**
Daily activities		
Nutrition	8.0 (1.5)	29%
Fitness	8.0 (1.3)	33%
Personal care	7.8 (1.4)	43%
Communication	8.2 (1.0)	26%
Housing	7.1 (1.2)	64%
Mobility	7.0 (1.3)	71%
Daily activities subdomain	***7.6 (1.0)***	***57%***
Social roles		
Responsibility	8.6 (0.9)	17%
Relationships	8.5 (0.9)	19%
Community life	8.1 (1.5)	24%
Education*	8.3 (1.0)	38%
Employment*	7.8 (1.3)	44%
Recreation	7.3 (1.7)	57%
Social roles subdomain	***8.0 (1.1)***	***36%***

* The category of education was applicable for 8 participants and the category of employment for 25 participants.

Participation

Mean scores on participation are presented in Table 2. On average, 57% of the participants had difficulties in daily activities. The most frequently cited difficulties were in the areas of mobility, housing and personal care. Thirty-six percent of the participants had difficulties in social roles; with recreation, employment and education cited as the most problematic areas.

Adjusting for gender and gross motor functioning, adults with a higher level of physical fitness had less difficulties in social roles (OR= 34.1, p= .03) and tended to have less difficulties in daily activities (OR= 9.3, p= .09). Level of everyday PA did not influence participation in the multivariate analyses (Table 3).

Health-Related Quality of Life

Mean scores on HRQoL are presented in Table 4. The mean score in the physical domain (PCS) was 46.7 (7.3) and in the mental domain (MCS) was 52.9 (10.4). The lowest HRQoL was experienced in the domains of vitality, physical functioning, and role limitation physical.

After adjusting for gender and gross motor functioning, adults with a higher level of physical fitness also had a higher level of physical HRQoL (OR= 24.9, p= .02), but not a higher level of mental HRQoL (Table 5). Level of everyday PA was not

Table 3. Relationships between participation, physical fitness and level of everyday physical activity in adults with ambulatory cerebral palsy.

	Daily activities						Social roles					
	Univariate			*Multivariate*[1]			*Univariate*			*Multivariate*[1]		
	OR	95% CI	R^2	OR	95% CI	R^2	OR	95% CI	R^2	OR	95% CI	R^2
Basic characteristics												
GMFCS level (III-IV, II, I)	**10.6****	**2.3-49.7**	**41%**	-	-	-	**9.1****	**2.0-41.4**	**37%**	-	-	-
Gender (male, female)	1.9	0.5-7.2	3%	-	-	-	1.4	0.3-5.6	1%	-	-	-
Lifestyle characteristics												
VO_{2peak} (L/min)	2.6	0.7-10.2	6%	**9.3**#	**0.7-125.9**	**49%**	**4.6***	**1.0-21.4**	**13%**	**34.1***	**1.4-870.7**	**52%**
Duration of dynamic activities, %	1.1	0.9-1.3	2%	0.9	0.7-1.2	42%	**1.3***	**1.0-1.6**	**15%**	1.1	0.9-1.4	40%

[1] Separate regression models were built for each factor (physical fitness and level of everyday PA), adjusted for GMFCS level and gender. Provided R^2 values represent the explained variance of the whole model.

* $p \leq .05$

** $p \leq .01$

\# $p < .10$

OR = Odds Ratio, CI = Confidence interval, R^2 = Nagelkerke R squared, represents explained variance

Table 4. Health-related quality of life in ambulatory adults with cerebral palsy.

	Mean (SD) (range 0-100)
Physical functioning	66.1 (21.6)
Role limitation physical	72.6 (35.3)
Bodily pain*	80.5 (21.2)
General health perception	76.8 (13.5)
Vitality	65.1 (17.3)
Social functioning	82.7 (19.1)
Role limitation emotional	83.3 (26.8)
Mental health	76.1 (17.6)
Physical component summary	**46.7 (7.3)**
Mental component summary	**52.9 (10.4)**

* *Two missing values on bodily pain*

related to either physical or mental HRQoL after adjusting for gender and gross motor functioning.

DISCUSSION

The results of the present study indicate that a higher level of physical fitness correlates to less difficulty in daily activities and social roles and to a better physical HRQoL. Similar results were obtained when analyses were carried out with physical fitness corrected for body mass (VO_{2peak} in mL/kg/min). We did not find evidence for an association between level of everyday PA and participation or HRQoL.

The most often cited difficulties were in the physical domain, in regard to both participation and HRQoL. More than half of the participants experienced difficulties in daily activities. Overall, HRQoL scores for vitality, physical functioning and role limitation physical were low. These results are in agreement with a former study in adolescents and young adults with CP.[13]

Participation

Physical fitness, but not level of everyday PA, was associated with difficulties in daily activities and social roles in the present study. A higher level of physical fitness may possibly result in lower levels of physical strain experienced during everyday activities, and thus to less difficulty in these daily activities and in social participation. Buffart and co-workers[10] showed a similar relationship between physical fitness and participation in young adults with myelomeningocele. However, physical fitness plays a less important role in participation in patient with spinal cord injury.[27] Together, these results may be due to the fact that CP and myelomeningocele are childhood-onset

Table 5. Relationships between health-related quality of life, physical fitness and level of everyday physical activity in adults with ambulatory cerebral palsy.

	Physical Component Summary						**Mental Component Summary**					
	Univariate			*Multivariate*[1]			*Univariate*			*Multivariate*[1]		
	OR	95%CI	R^2	OR	95%CI	R^2	OR	95%CI	R^2	OR	95%CI	R^2
Basic characteristics												
GMFCS level (III-IV, II, I)	2.4	0.8-6.8	9%	-	-	-	0.7	0.2-1.9	2%	-	-	-
Gender (male, female)	1.3	0.3-4.9	0%	-	-	-	3.2	0.6-17.7	7%	-	-	-
Lifestyle characteristics												
VO_{2peak} (L/min)	**4.3**$^{\#}$	**0.9-19.7**	**13%**	**24.9***	**1.8-3.6**	**30%**	0.5	0.1-2.3	3%	2.1	0.2-20.4	12%
Duration of dynamic activities, %	**1.2**$^{\#}$	**1.0-1.4**	**10%**	1.1	0.9-1.4	14%	1.0	0.8-1.2	1%	1.0	0.8-1.2	11%
Participation												
Daily activities, % difficulty	1.6	0.4-5.9	2%	0.8	0.2-4.1	9%	1.1	0.3-4.1	0%	1.4	0.3-7.3	11%
Social roles, % difficulty	1.0	0.2-3.7	0%	0.4	0.1-2.1	13%	1.5	0.4-6.1	1%	2.5	0.5-18.0	15%

[1] Separate regression models were built for each factor (physical fitness and level of everyday PA), adjusted for GMFCS level and gender. Provided R^2 values represent the explained variance of the whole model.

* $p \leq .05$

** $p \leq .01$

$^{\#}$ $p < .10$

OR = Odds Ratio, CI = Confidence interval, R^2 = Nagelkerke R squared, represents explained variance conditions, while spinal cord injury often occurs at a later age, which may coincide with different standards and internal evaluation of participation.

The absence of a relationship between the level of everyday PA and participation was contrary to what we expected. We expected the level of everyday PA to be correlated primarily to participation in daily activities. It is possible that the variation in level of everyday PA was not great enough to have strong effects on participation in daily activities and social roles. In addition, everyday PA is a behavior related to the movements that people perform, and the level of everyday PA may not have been high enough to relate to difficulty in participation as assessed with the Life habits questionnaire. In adults with unilateral CP, only participation in sports was associated with level of everyday PA, while other domains of participation, such as work were not associated.[8] In other groups such as patients with myelomeningocele and spinal cord injury, the level of everyday PA did play a role in participation.[10,27] However, the variation in levels of everyday PA was greater in these studies than that found here.

Health-Related Quality of Life

The current results underline the importance of physical fitness to the physical aspect of HRQoL. However, due to the cross-sectional and correlational design of this study cause and effect cannot be concluded. Nevertheless, it is an important finding since physical fitness is a parameter which can be influenced by factors such as physical training. Previous studies on relationships between lifestyle and HRQoL in CP used PA as a measure for lifestyle[9,15], yielding similar results to the present study in that only a weak relationship between PA and HRQoL was found.[9] Another study investigated the relationship between PA and self-reported quality of life in ambulatory adolescents with CP.[15] Measurements of PA in terms of walking activity (number of steps) and a questionnaire concerning activities in the past seven days, showed no relationship to self-reported quality of life.

The mental aspect of HRQoL was not associated with either physical fitness or PA. Variables other than lifestyle factors may play a role in mental well-being, such as self-efficacy, coping strategies, personality, educational level or social support.[9,12,14]

The current study emphasizes the importance of physical fitness in participation in daily activities and social roles and in physical HRQoL. Future research is warranted to determine the effect of interventions based on physical fitness in order to improve participation in daily activities and physical HRQoL in persons with CP.

Limitations of the study

Generalization of our results is limited to ambulatory adults with bilateral spastic CP, due to inclusion criteria for cycle ergometer testing and the exclusion of persons with CP dependent on electric wheelchairs. Different results may be obtained in a population of adults with more severe CP in terms of gross motor functioning. Furthermore, general questionnaires, not specifically designed for persons with CP were used in the present study. The SF-36 may not have been sensitive enough for condition-specific aspects of HRQoL, which may have influenced the amount and the magnitude of the relationships found. However, the advantage of using generic questionnaires is that comparisons between different diagnosis groups are possible.

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CHAPTER 5

Experienced problems of young adults with cerebral palsy: targets for rehabilitation care

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ABSTRACT

Objective To determine the problems experienced by young adults with cerebral palsy (CP) and the relationship between those problems and personal and CP-related characteristics.

Methods A cross-sectional study was performed in young adults (*n*=87; aged 18-22y) with CP and normal intelligence (roughly corresponding to an intelligence quotient >70, excluding participants who attended schools for those with learning disabilities). We used the Canadian Occupational Performance Measure in a semistructured interview to assess participants for experienced problems. We further categorized experienced problems according to the domains of the *International Classification of Functioning, Disability and Health* and assessed the relationship between those problems and personal and CP-related characteristics (e.g. age, sex, level of gross motor functioning, manual ability, level of education) using appropriate correlation coefficients.

Results Approximately 70% of participants experienced problems in daily life, addressing the areas of self-care (59%), productivity (52%) and leisure activities (37%). More specifically, problems were most prevalent in recreation and leisure (30%), preparing meals (29%), housework (14%) and dressing (14%). Problems in functional mobility, paid or unpaid work, and socialization were considered as most important (represented by the highest mean importance score). Mobility problems were associated with lower levels of gross motor functioning (Spearman ρ= 0.39), and problems with self-care were associated with lower levels of manual ability (Spearman ρ= 0.40).

Conclusions Although frequently addressed during pediatric rehabilitation care, problems with mobility and self-care still prevail in young adults with CP. In addition, during the transition into adulthood young adults with CP may experience problems regarding domestic life and work, which they consider important.

INTRODUCTION

Cerebral palsy (CP) is one of the most frequently occurring medical conditions of childhood.[1-3] In the Netherlands, 1.51 per 1000 inhabitants have CP, and epidemiologic data suggest that this prevalence has not decreased over time.[4] Children with CP account for approximately 50% of pediatric rehabilitation patients in the Netherlands, and this population has received much attention in pediatric rehabilitation medicine regarding the provision of health care services.[5] However, as they enter adulthood, attention for young adults with CP seriously declines, despite the physical, social and emotional changes that may occur during this transition.[6]

There has been a recent increased awareness about the need for lifelong care of persons with CP.[7,8] It is recognized that young adults with childhood disability might encounter difficulties during their transition into adulthood that require treatment and care.[9-11] The lack of coordinated services for these young adults with CP has prompted several researchers to propose a multidisciplinary approach to care.[6,12-15] In the United Kingdom, specialized young adult teams were designed to improve the transition from child to adult care; evaluations of these interventions show that they lead to improved participation in society for young adults with CP.[6,7] Young adult teams are also available in Sweden[16], and the first Dutch young adult team started in 2007.[17,18]

Although the number of lifespan care initiatives is growing, we have limited systematic knowledge of specific problems encountered by young adults with CP during the transition to adulthood. In a previous study with young adults with CP[19], we demonstrated several unmet needs, primarily involving information, mobility and health services. Although young adults with low levels of gross motor functioning reported the most unmet needs, those with higher functioning also reported several unmet needs.[19] Donkervoort et al.[20] reported that 20% to 30% of young adults with CP encountered difficulties in performing daily activities and social participation. They experienced difficulties mainly in mobility, self care, nutrition, employment and community life.[20,21] Studies of adults with CP have also shown that limitations in activities of daily life are present in several areas such as self-care and mobility.[22-24]

Further knowledge of experienced problems of young adults themselves is of great value in determining relevant topics at which the young adult teams should be aimed. Therefore, the current study aimed to determine the experienced problems of young adults with CP of normal intelligence, using a client-centered perspective.

METHODS

Study sample

This study is part of the CP Transition Study Southwest Netherlands, which was designed to evaluate the course of functioning in adolescents and young adults with CP over time.[20,21] Data described in the current study were collected at the 2-year follow-up, when the young adults were aged 18 to 22 years. Participants were recruited from 8 participating rehabilitation centers and rehabilitation departments in the southwest area of the Netherlands. The inclusion criteria for the original study[20,21] were (1) diagnosis of CP, (2) age 16 to 20 years and (3) normal intelligence (excluding participants who attended schools for those with learning disabilities; roughly corresponding to an intelligence quotient >70). Exclusion criteria were (1) secondary medical conditions having lasting effects on motor functioning and (2) insufficient knowledge of the Dutch language. Participants received verbal and written information about the study and gave verbal and written consent to participate. Ethical approval for the study was provided by the Medical Ethics Committee of Erasmus Medical Center, Rotterdam.

In total, 87 of 103 participants from the original study participated in the 2-year follow-up (dropout rate, 16%). Dropout occurred secondary to loss of interest (n=9), competing time demands (n=5) or relocation to another country (n=2). When we compared baseline characteristics of participants and dropouts (n=16), no significant differences were found in sex, age, gross motor function, manual ability, type of CP, limb distribution or housing status. Persons who dropped out of the study had a lower educational level (39% versus 27% with low level of education; see below for description of educational levels) compared to participants (p= .02).

Measurements

Three interviewers who were trained in the Canadian Occupational Performance Measure (COPM) assessed problems experienced by the participants with the COPM, which is a semistructured interview tool.[25] We used the structure of the *International Classification of Functioning, Disability and Health* (ICF)[26] to further organize and link experienced problems. Linking the problems to the ICF may allow standardization and comparability to other studies because of this tool's widespread use. These linking rules have been applied in previous studies to compare measurement instruments[27-29] and for goal-setting in rehabilitation care.[30,31]

The interviewer noted personal and CP-related characteristics, including sex, age, limb distribution of the paresis, level of gross motor functioning (Gross Motor Functioning Classification System (GMFCS))[32] and manual ability (Manual Ability Classification System (MACS)).[33] The GMFCS is a 5-level classification system of gross motor functional abilities and limitations; a higher GMFCS level (level V) corresponds to a lower level of gross motor functioning.[32] The MACS is a 5-level classification tool used to evaluate how persons with CP handle objects during daily activities; a higher MACS level (MACS level V) corresponds to a lower level of manual ability.[33] Both the GMFCS and MACS were originally developed and validated for children[32,33], but have demonstrated reliability and validity in young adults.[34,35]

We classified participants by 3 different educational levels based on actual and potential educational capacity: low (prevocational practical education or less), medium (prevocational theoretical education and upper secondary vocational education), or high (secondary non-vocational education, higher education and university).[20]

Canadian Occupational Performance Measure

The COPM[24] is a semistructured interview tool that focuses on activities that the participant wants, needs, or is expected to perform. The COPM was developed to assess experienced problems in 3 areas of occupational performance: self-care, productivity, and leisure. Each performance area in turn covers 3 activity domains. Within an activity domain the participant can report several experienced problems. The importance of each problem, as perceived by the participant, is first rated on a 10-point scale, with 1 being not important and 10 being extremely important. Participants then name the 5 activities that are most important to them. The COPM also assesses activity performance and satisfaction with performance, but these areas will not be discussed here. Several studies have shown that the COPM has good validity, moderate test-retest and inter-rater reliability, and is sensitive to change.[36-40]

Types of problems: International Classification of Functioning, Disability and Health Linking

To further categorize specific types of problems experienced by young adults with CP in a more standardized way, we linked the experienced problems to the ICF. The ICF[26] provides a framework for recording and organizing information about health and health-related states in a standardized, common language that facilitates communication between various disciplines and scientific fields.[41,42] The ICF assesses: (1) functioning and disability, and (2) contextual factors. The COPM is an instrument focussing on activities, so contextual factors (part II) were not considered in this study. Part I includes the components *body functions* (“b”), *body structures* (“s”), and *activities* and *participation* (“d”). In the ICF classification, “b”, “s” and “d” are followed by numeric codes starting with the chapter number (domain, 1 digit), followed by the second level (2 digits), third level (1 digit) and fourth level (1 digit), which are the categories of the ICF. For example, “hand or arm use” is coded as d440, where “d” indicates the component activities and participation, the first “4” indicates the mobility domain, and “40” indicates the second level category of hand or arm use.

In this study, we linked each problem reported on the COPM to the most precise ICF category, by applying the linking rules of Cieza et al.[41,43] Two researchers (C.N., M.D.) determined to which ICF category the experienced problems should be linked. In cases of disagreement, a third researcher (M.E.R.) was consulted. In this article, results of second level linking are presented.

Data analysis

Results were summarized using descriptive statistics. Associations between experienced problems and personal and CP-related characteristics (e.g. age, sex, level of gross motor functioning, manual ability, level of education) were determined with appropriate correlation coefficients: Phi coefficients *(Phi)* for 2 dichotomous variables, Pearson correlation coefficients *(Rp)* for associations between dichotomous and continuous variables and Spearman correlation coefficients *(ρ)* for associations between dichotomous and ordinal variables. Because of the large number of relationships evaluated, a significance level of .01 or less was used.[44] According to this level of significance and available sample size, the power of the study was 0.90 to detect associations of 0.40 or greater. All statistics were performed using Statistical Package for the Social Sciences (Windows version 12.0.1).[a]

RESULTS

Study sample

Participant characteristics are presented in Table 1. The study sample included 87 young adults (51 men, 36 women) with CP and normal intelligence. Their mean age ± SD was 19.9±1.4 years. Approximately half of the participants had hemiplegic CP, 31% had diplegic CP, and 20% had quadriplegic CP. Most participants had a high level of gross motor functioning (i.e. they could climb stairs, had no limitations in walking indoors or outdoors), and had a high level of manual ability. Participants were evenly distributed across educational levels, with 40% at the middle educational level (prevocational theoretical education and upper secondary vocational education).

Experienced problems

Participants reported a total of 271 experienced problems, with the number of problems per person ranging from 0 to 15. Approximately 29% of participants reported no experienced problems on the COPM. At least 1 problem was reported by 51 participants (59%) for self-care, 45 participants (52%) for productivity, and 32 participants

Table 1. Participant characteristics.

	Participants (*n*=87)
Age (y)	19.9±1.4
Sex (men)	51 (59)
Limb distribution	
Hemiplegia	43 (49)
Diplegia	27 (31)
Quadriplegia	17(20)
Type of CP*	
Spastic	82 (94)
Ataxic	1 (1)
Dyskinetic	1 (1)
Mixed	2 (2)
GMFCS level*	
I	63 (72)
II	9 (10)
III	5 (6)
IV	8 (9)
V	1 (1)
MACS level*	
I	67 (77)
II	10 (12)
III	4 (5)
IV	1 (1)
V	1 (1)
Educational level	
Low	23 (27)
Middle	35 (40)
High	29 (33)
Student/employment	
Student	53 (61)
Paid job	17 (20)
Other (e.g. day center)	17 (19)
Housing status*	
Living with parents	61 (70)
Living alone or with others	19 (22)
Living in an institute	6 (8)

NOTE. Values are mean±SD or *n* (%).

* Missing data for type of CP (*n*=1), GMFCS (*n*=1), MACS (*n*=4) and housing status (*n*=1)

(37%) for leisure (Table 2). Commonly reported problems were cutting nails, cutting food into pieces, and walking long distances.

Table 2 also shows the prioritization of experienced problems in the different COPM domains. Young adults with CP identified the following as priority problem areas (proportion of participants reporting at least 1 problem): functional mobility (39%), paid or unpaid work (24%), household management (8%) and personal care (8%). Although not mentioned often as being a number 1 priority, the mean importance rating of problems with socialization was high (8.8±1.2). Other domains with high

Table 2. Experienced problems, Priorities, and Importance Ratings, as reported on the COPM.

	No. of participants reporting a problem		**No 1. priority***		**Mean importance of problems (0-10)**	**Most often mentioned problems**
Self care	**51**	**(59)**				
Personal care	22	(25)	5	(8)	6.7±2.5	Cutting nails, buttoning shirt, tying shoelaces
Functional mobility	41	(47)	24	(39)	7.6±2.3	Walking long distances, standing for a long time, walking stairs
Community management	10	(12)	4	(6)	7.8±1.5	Purchasing groceries/carrying bags with groceries
Productivity	**45**	**(52)**				
Paid or unpaid work	22	(25)	15	(24)	8.7±1.3	Finding a job/other employment, having fun in employment
Household management	29	(33)	5	(8)	7.1±2.1	Cutting food into pieces, peeling potatoes, cooking
Play/school	11	(13)	3	(5)	7.8±1.6	Writing
Leisure	**32**	**(37)**				
Quiet recreation	5	(6)	0	(0)	6.4±2.0	Playing the guitar
Active recreation	24	(28)	4	(6)	6.0±2.6	Playing soccer, ice-skating, horseback riding
Socialization	9	(10)	2	(3)	8.8± 1.2	Contact with non-familiar persons, going out
No problems reported	**25**	**(29)**	-		-	-

NOTE. Values are *n*(%) or mean ± SD.

* Percentages were calculated for persons who reported at least 1 problem (*n*=62).

mean importance ratings were paid or unpaid work (8.7±1.3), community management (7.8±1.5) and play/school (7.8±1.6).

Types of problems

We linked 98% of experienced problems detected by the COPM to the activity and participation components of the ICF. The remaining 2% (5 problems) were linked to the body functions component. Table 3 shows the ICF categories for which at least 10% of participants reported a problem. Problems were experienced in ICF categories of recreation and leisure (30%), preparing meals (29%), doing housework (14%) and dressing (14%). Other self-care activities, work, and specific mobility problems were also reported as experienced problems.

Relationships

The presence of a problem in different domains of the COPM was not associated with sex or age (Table 4). Participants with lower levels of gross motor functioning (GMFCS level) experienced more problems in functional mobility (Spearman $\rho= 0.39$, $p< .001$) and in socialization (Spearman $\rho= 0.40$, $p< .001$). Persons with

Table 3. Types of problems experienced by more than 10% of young adults with cerebral palsy, as classified by the ICF.

	ICF code	No. of participants reporting a problem	
		n	(%)
Recreation and leisure	d920	26	(30)
Sports			
Socializing			
Arts and culture			
Crafts			
Recreation and leisure, unspecified			
Preparing meals	d630	25	(29)
Preparing simple meals			
Preparing meals, unspecified			
Preparing complex meals			
Doing housework	d640	12	(14)
Using household appliances			
Cleaning cooking area and utensils			
Cleaning living area			
Doing housework, other specified			
Doing housework, unspecified			
Washing and drying clothes and garments			
Dressing	d540	12	(14)
Putting on clothes			
Putting on footwear			
Caring for body parts	d520	11	(13)
Caring for fingernails			
Caring for hair			
Caring for skin			
Caring for teeth			
Acquiring, keeping and terminating a job	d845	11	(13)
Seeking employment			
Walking	d450	11	(13)
Walking long distances			
Walking on different surfaces			
Walking around obstacles			
Walking, other specified			
Lifting and carrying objects	d430	11	(13)
Carrying in the hands			
Lifting			
Using transportation	d470	11	(13)
Using private motorized transportation			
Using public motorized transportation			
Using transportation, unspecified			
Driving	d475	9	(10)
Driving motorized vehicles			
Driving human-powered transportation			

NOTE. Values are *n*(%) or as otherwise indicated.

lower levels of manual ability (MACS level) experienced more problems in personal care (Spearman ρ= 0.32, p= .003) and in play/school (Spearman ρ= 0.29, p= .008). Finally, those with lower levels of education, experienced more problems in (un) paid work (Spearman ρ= -0.30, p= .005).

Table 4. Associations between personal and cerebral palsy-related characteristics and experienced problems.

COPM domain	Sex[*]	Age[†]	GMFCS level[‡]	MACS level[‡]	Level of education[‡, §]
Personal care	-0.01	-0.24	0.06	0.32[‖]	-0.12
Functional mobility	0.14	0.16	0.38[‖]	0.17	0.07
Community management	0.21	0.05	0.15	-0.05	-0.26
(Un) paid work	0.05	-0.02	0.10	0.07	-0.30[‖]
Household activities	0.10	-0.14	0.03	0.11	-0.13
Play/school	0.03	-0.10	0.12	0.29[‖]	-0.04
Quiet recreation	-0.11	-0.05	-0.15	0.02	0.11
Active recreation	0.21	0.06	-0.08	-0.09	0.08
Socialization	0.02	-0.06	0.38[‖]	0.15	-0.17

[*] Phi coefficient was calculated for dichotomous variables.

[†] Pearson correlation coefficients for continuous variables.

[‡] Spearman correlation coefficients for ordinal variables.

[§] Level of education was classified as low (prevocational practical education or less), medium (prevocational theoretical education and upper secondary vocational education), or high (secondary non-vocational education, higher education, and university).

[‖] $p \leq .01$.

DISCUSSION

This is the first study that used the client-centered perspective to determine the problems that young adults with CP experience. We showed that young adults with CP with normal intelligence experienced several problems in daily life. Problem areas primarily include mobility, work, self-care, domestic tasks and leisure. Persons with lower level levels of gross motor functioning and lower levels of manual ability appear to be more prone to experiencing problems in daily life.

Mobility

Problems in mobility are reported frequently and are of high importance for young adults with CP. So although much attention is given to problems in mobility in pediatric rehabilitation, it appears that these problems will remain important throughout adulthood. Partly, these problems address walking and wheelchair use, which might be considered to be inherent to lower levels of gross motor functioning in CP. But they also refer to problems in using public transportation or driving a car, which are considered more important at adult age. The reporting of more mobility problems in young adults with lower levels of gross motor functioning is consistent with other studies of children and young adults with CP.[1,20,45]

Other studies confirm our results regarding the persisting presence of mobility issues In young adults with CP.[19,21,46] We previously showed that 66% of young adults with CP with normal intelligence experienced unmet mobility needs.[19] Donkervoort et al.[21] recently reported that 31% of participants experienced mobility difficulties related to daily activities and social participation (as measured by the Life-Habits questionnaire). Bax et al.[46] found that young adults with CP with varying levels of physical and cognitive impairments became less mobile after leaving school and that many did not receive further medical examinations to assess their physical condition after leaving school. Also at adult age, persons with CP reported several types of problems with mobility.[22,47,48] Andersson et al.[47] found that nearly half of the adults with CP without learning disabilities could not move independently within the community as desired or required.

Work

Problems in work were experienced less often compared with other domains, but most participants who did mention problems rated them as very important. The small number of participants reporting work problems may reflect the young age of the participants and that they may still be in school rather than working. These problems arose mainly in participants with lower levels of education, who are the ones who had finished their education and are now working or looking for work. Also, other studies report work problems in young adults with CP.[21,49,50] Therefore, work problems are important to recognize in young adults with CP.

Self-care, domestic tasks and leisure

Although self-care is often addressed in pediatric rehabilitation, apparently in young adults with CP problems are still present in this domain. Our results are consistent with previously published studies that show that young adults with CP experience difficulties with daily activities and participation in personal care (21%).[21] Studies of adults with CP also show that the execution of self-care tasks (e.g. bathing and dressing) poses difficulties.[48,51] Because self-care activities depend primarily on the use of the upper extremities, participants with more impaired manual ability (as determined by MACS level) are probably more prone to problems in these activities.

Problems with the performance of domestic tasks are rarely discussed in the literature. Andren et al.[48] reported that persons with inborn or early-acquired mobility disorders (most of whom had CP), demonstrated the highest level of dependence on aids or other persons for activities related to household tasks. Several of our study respondents indicated that performing domestic tasks was too physically demanding because they needed to conserve energy for work, education or mobility-related activities.

For leisure activities most participants indicated problems with physically demanding activities (e.g. team sports). This is in line with the experienced problems in mobility. No relationships were found with specific personal or CP-related characteristics.

The process of linking

The process of linking experienced problems on the COPM to ICF domains was easy and provides a way to further categorize problems that young adults with CP experience in daily life. Describing these problems in terms of the ICF might increase the comparability of studies into the experienced problems of young people with disabilities in which different assessment methods were used.[29,43]

Limitations

Our study findings may be limited in their generalizability to other countries than the Netherlands and to other CP populations. This study focused on young adults with CP without learning disabilities, whom we expect to be capable of living an independent adult life. The selection criteria produced a cohort of well-functioning young adults, about three quarters of whom functioned at the GMFCS level I and MACS level I. Consequently, generalization of our results is limited to those young adults with CP with normal intelligence and to those with rather high levels of gross motor functioning and manual ability. Also, the moderate associations between personal and CP-related characteristics and experienced problems should be interpreted cautiously.

Implications

When growing into adulthood, young adults with disabilities have to learn a range of life skills that enable them to participate in adult life, such as finding a vocation, traveling to work and doing housework.[7] The current study showed that young adults with CP rated problems in participation as very important. Although topics such as mobility and self-care are frequently addressed in pediatric rehabilitation interventions, problems in these areas might still exist during young adulthood. Furthermore, problems with regard to work and domestic life may appear when the child with CP grows older, which can be indicated as growing into a deficit.[21] In this sample with predominantly young adults with high levels of gross motor functioning, mobility still is an important issue. Although the general beliefs may be that those with higher levels of gross motor functioning have little problems in mobility, this study points in another direction. Other problems in mobility may arise when using transportation for work or in recreation such as going out to a bar (standing for a long time).

The problems that young adults with CP perceive indicate the need for them to acquire life skills that will enable them to participate in adult life.[7] Therapists and physicians should be aware that new types of problems may arise when a young adult with CP reaches adulthood, which may warrant another approach or interventions than in pediatric care. It is important to consider patient experiences and priorities in setting treatment goals for young adults with CP.

We used the present study results to develop age-appropriate interventions (in addition to regular rehabilitation appointments) for young adults to address specific areas of participation: work[52], household chorus, directing one's own life, pursuing an active lifestyle, and sports participation.[18] These are now offered as part of a modular treatment program in a new young adults teams program in the Netherlands.[17,18] Further evaluation of these interventions is needed to improve rehabilitation care for young adults with CP.

CONCLUSIONS

Young adults with CP with normal intelligence experienced several problems in daily life. Although frequently addressed during pediatric rehabilitation care, problems with mobility and self-care still prevail in young adults with CP. In addition, during the transition into adulthood young adults with CP may experience problems regarding domestic life and work, which they consider important.

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CHAPTER 6

Unmet needs and health care utilization in young adults with cerebral palsy

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ABSTRACT

Objective To gain insight into the unmet needs and utilization of health care of young adults with cerebral palsy (CP) and to explore relations between unmet needs, health care utilization and subject characteristics.

Methods A cross-sectional study was performed in 29 young adults with CP without severe learning disabilities (IQ>70). Subject characteristics such as age, gender, limb distribution, level of gross motor functioning, level of education and perceived participation and autonomy were measured. Outcome measures were the Southampton Needs Assessment Questionnaire, Impact on Participation and Autonomy and a questionnaire on health care utilization.

Results Young adults with CP reported unmet needs mostly on information (79%), mobility (66%) and health care (66%). About half of the participants visited a rehabilitation physician (52%) or a physical therapist (55%) in the past year. Participants with lower levels of gross motor functioning were found to have more unmet needs and visited various health care professionals more often than young adults with higher levels of gross motor functioning. However, participants with higher levels of gross motor functioning still reported several unmet needs.

Conclusions Although young adults with CP frequently receive treatment from health care professionals, they indicate unmet needs with respect to several areas such as information on diagnosis, functional mobility and formal health care. In the treatment of young adults with CP, attention should be paid to these aspects.

INTRODUCTION

Cerebral palsy (CP), defined as an umbrella term covering a group of non-progressive (but often changing) motor impairment syndromes secondary to lesions or anomalies of the brain arising in the early stages of development[1], is one of the most frequently occurring conditions in childhood.[2-4] Since about 50% of the paediatric rehabilitation population in the Netherlands consists of children with CP, this group of children has received much attention in paediatric rehabilitation medicine.[5] However, after discharge from paediatric rehabilitation, attention for this group seriously declines. This decline takes place just in a period of significant physical, emotional and social changes.[6]

For adolescents with physical disabilities, the transition from childhood to adulthood may be more complicated as their physical limitations may restrict their pursuit of independence and the formation of social relationships outside the family.[7,8] Health care professionals could serve as a consultant in the transition process of adolescence to adulthood to promote a smooth transition.[6,7,9-11]

Problems that young adults with CP encounter are different from those they experienced in childhood, which might result in new unmet needs for which further treatment may be warranted.[12] However, several studies reported a discontinuity of care when young adults with physical disabilities enter adulthood and reported diminished contact with health care after leaving school.[13-17] Utilization of health care among school leavers declined significantly after leaving school in terms of both medical specialist care (e.g. rehabilitation physician, neurologist) and therapy (e.g. physical therapist, occupational therapist). Both the number of young adults visiting a particular health care practitioner as well as the frequency of contacts tends to diminish after leaving school.[14,15,17]

A lack of adequate care, together with changes in social role and in environmental expectations as they grow into adulthood, may result in unmet (health) needs of young adults with physical disabilities.[13,14,18] It remains unclear if unmet needs are present in young adults with CP and, in case they do exist, which ones they are. It is important to know what the unmet needs of young adults with CP are in order to improve the continuity of care and to effectuate an adequate hand-over to adult services after discharge from paediatric rehabilitation. The aim of the current study is (*i*) to obtain insight in unmet needs and health care utilization of young adults with CP, and (*ii*) to explore relations between unmet needs, health care utilization and subject characteristics, in the search for specific subgroups of patients with many needs and high utilization of health care.

METHODS

Sample

Inclusion criteria for the current study were diagnosis of CP, aged between 16 and 40 years and known to the paediatric or adult department of a rehabilitation centre in two cities in the South-West Netherlands (Rotterdam and The Hague). In the Netherlands, the vast majority of children with CP is treated in a rehabilitation centre. These rehabilitation centres have a regional function, covering these cities and their surroundings.

Young adults with severe learning disabilities (IQ <70), inadequate language comprehension to fill out the questionnaires, and those with additional diagnosis with lasting effects on motor functioning were excluded. Of 64 eligible participants, 29 persons agreed to participate (response rate 45%). No significant differences were found between responders and non-responders with regard to age, gender, level of gross motor functioning, limb distribution of the paresis and level of education (Table I). Participants received written information and gave written consent to participate.

Measurement procedure

Several subject characteristics were determined, e.g. age, gender, level of gross motor functioning, limb distribution, level of education, employment status, housing status, and perceived participation and autonomy. Postal questionnaires on unmet needs and health care utilization were sent to all eligible subjects. After approximately six weeks, a reminder letter was sent to non-responders.

Measurements

Subject characteristics

The level of gross motor functioning was determined using the Gross Motor Functioning Classification System (GMFCS), a classification system for persons with CP which is based on spontaneous movements with regard to sitting and walking.[19] The GMFCS consists of five levels ranging from “walks without restrictions” (level I) to “self-mobility is severely limited even with use of assistive technology” (level V). Distinction between the five different levels is focused on functional limitations and need for assistive technology, including mobility devices and wheeled mobility rather than quality of movement. Inter-rater reliability and content and predictive validity have been established for children.[19]

Level of education was subdivided in three levels, referring to actual and potential educational capacity: low, including prevocational practical education or less; medium, including prevocational theoretical education and upper secondary vocational education; high, including secondary non-vocational education, higher education and university. Perceived participation and autonomy was determined using the first part of the Impact on Participation and Autonomy (IPA) (see below). [20,21]

Unmet needs

No specific instruments were available for measurement of the unmet needs of young adults with CP. Therefore, to study these needs we used part of a generic instrument for adults, the IPA[20,21] and we adapted a needs assessment questionnaire for stroke patients, i.e. the Southampton Needs Assessment Questionnaire (stroke version) (SNAQ) for young adults with CP (Kersten, pers. communication).[22-24]

IPA

The IPA is a generic measure, addressing the impact of a chronic condition on a person's participation and autonomy (31 items) and related problem-experience (eight items).[20,21] The IPA consists of five participation domains: autonomy indoors (seven items, e.g. getting around at home, self-care activities), family role (seven items, e.g. looking after home, economic self-sufficiency), autonomy outdoors (five items, e.g. visiting friends, spending leisure time), social relations (six items, e.g. quality of relationships, receiving respect) and work & education (six items, e.g. doing the desired work or following the desired education). Perceived participation and autonomy is reflected on a 0-4 scale: very good (0), good (1), fair (2), poor (3), very poor (4). Standardized mean scores are calculated by dividing the mean domain scores by the number of items in that domain. A higher score represents greater restrictions in participation and autonomy. In the current study, a score of 3 or 4 was considered to indicate restricted participation.

Problem-experience is determined on eight items: mobility, self-care, family role, finances, leisure, social relations, occupation, education. In addition to the original IPA, for these eight items a question on need for support and/or adaptations was added. Both problem-experience and need for support and/or adaptations are reflected on a 0-2 scale: no problems (0), minor problems (1), severe problems (2), and support and/or adaptations are adequate (0), need less support and/or adaptations (1), need more support and/or adaptations (2), respectively. Higher scores represent greater problem-experience.

The original IPA has been shown to have good test-retest reliability, good convergent and divergent validity, and has shown to be responsive to change in several diagnostic groups such as stroke, spinal cord injury and rheumatoid arthritis.[25,26]

SNAQ

The SNAQ is an instrument which can be used to describe the unmet needs of patients in several diagnostic groups.[22-24] In the current study, we adapted the SNAQs (originally developed for patients with stroke) for young adults with CP in cooperation with the original authors of the SNAQ and translated the questionnaire into Dutch. The modified SNAQ used in this study consists of 71 items divided over 10 domains (information, mobility, social activities, work or training, family, voluntary organizations, finances, housing, formal health services, formal social services). Possible responses are yes, no and not applicable. To make comparisons between the different need domains, standardized domain scores are calculated for each domain by dividing the number of reported unmet needs by the maximal score on the domain concerned (domain scores ranging from 0 – 100). Higher scores indicate more unmet needs.

The SNAQ has been shown to have good validity (content, coverage and construct), internal reliability, sensitivity and responsiveness for patients with different diagnoses.[24]

Health care utilization

Health care utilization is measured by questioning the participants on the number of contacts they had in the past year with therapists (e.g. physical therapist, speech therapist) and medical specialists (e.g. rehabilitation physician, orthopaedist). Possible responses are no contact, some contact, and regular contact (regular contact was defined as more than two times per month for therapists and more than two times per year for medical specialists). Furthermore, additional information is obtained concerning medication used and interventions.

Data analysis

Results on the IPA, SNAQ and on health care utilization are summarised using descriptive statistics. To study the relations between unmet needs (represented by domain scores on the SNAQ), health care utilization and subject characteristics, Spearman correlation coefficients are calculated. Differences between subgroups are studied using Mann Whitney U-tests. All statistics are done with Statistical Package for the Social Sciences (Windows version 12.0.1. SPSS Inc., Chicago, USA).

Table 1. Subject characteristics.

	Responders (*n*=29)		Non-responders[a] (*n*=35)	
Age in years, mean (SD)	28.1	(8.0)	28.1	(7.1)
Gender, *n* (%)				
Male	19	(66)	15	(43)
Female	10	(34)	20	(57)
GMFCS, *n* (%)				
I	15	(52)	13	(37)
II	4	(14)	10	(29)
III	3	(10)	1	(3)
IV	7	(24)	9	(26)
V	0	(0)	0	(0)
Limb distribution, *n* (%)				
Hemiplegia	8	(28)	10	(29)
Diplegia	7	(24)	11	(31)
Quadriplegia	14	(48)	12	(34)
Level of education, *n* (%)				
Low	14	(48)	13	(37)
Medium	8	(28)	11	(31)
High	7	(24)	5	(14)
Student/employment, *n* (%)				
Student	9	(31)	n.a.	
Regular employment	7	(24)	n.a.	
Other (e.g. day centre)	13	(43)	n.a.	
Housing status, *n* (%)				
Living with parents	11	(38)	n.a.	
Living alone or with others	11	(38)	n.a.	
Living in an institute	7	(24)	n.a.	

GMFCS, Gross Motor Functioning Classification System

[a] Missing data on GMFCS (*n*=2), limb distribution (*n*=2) and level of education (*n*=6). n.a. not assessed.

RESULTS

Participants

Subject characteristics (*n*=29) are presented in Table I. The study sample consisted of 19 male and 10 female young adults. Almost half of the participants were quadriplegic, a quarter hemiplegic and another quarter diplegic. The majority of the participants had a high level of gross motor functioning. With regard to level of education, almost half of the participants had a low level of education. At the time of the study, about one third of the participants was a student and the remaining participants had some kind of employment or other day occupation, such as attending an activity centre or voluntary work. Three quarters of the study sample either lived with their parents, alone or with others and one quarter lived in an institute.

With regard to their participation and autonomy, the young adults with CP in the current study felt most restricted in the areas of work and education (31%), autonomy outdoors (31%) and family role (38%) (Table 2).

Table 2. Participation and autonomy of young adults with cerebral palsy (IPA).

Domain	*Standardized mean score (sd)*	*Range (0-4)*	*Participants with restricted participation[a] (%)*	
Autonomy indoors	0.6 (0.5)	0 – 2.0	2	(7)
Family role	1.2 (0.7)	0 – 2.7	11	(38)
Autonomy outdoors	1.2 (0.9)	0 – 3.6	9	(31)
Social relations	0.9 (0.6)	0 – 1.8	7	(24)
Work & education[b]	1.2 (0.9)	0 – 3.4	9	(31)

IPA, Impact on Participation and Autonomy.

[a] Defined as a standardized mean score of 3 or 4

[b] Missing data (*n*=3) and only applicable for 18 participants.

Unmet needs

The percentage of participants experiencing problems in several domains of participation on the IPA are shown in Figure 1a. Young adults with CP experienced problems mostly in mobility (52% of the participants), followed by the other domains (27% to 43% of the participants indicated problems). The percentage of people expressing a need for more support and/or adaptations in these domains was small, ranging from six participants expressing a need on mobility (21%) to one participant expressing a need on self-care (3%).

Overall, young adults with CP reported a mean number of 11.9 (± 10.3) unmet needs on the domains of the SNAQ. More than three quarters of the participants (79%) reported at least one unmet need for information (e.g. information on complications of CP, on consequences of CP and on causes of CP) and two-third (66%) of the participants reported at least one unmet need on mobility (regarding access to the community, attitudes of other people and driving a car) and formal health services (e.g. physical therapy, in-patient rehabilitation and a hospital specialist), see Figure 1b. The numbers of unmet needs reported on each domain are given in Table 3. We found that most unmet needs were reported on the information domain, followed by unmet needs with regard to finances and social activities.

Relations were found between subject characteristics and number of unmet needs on the SNAQ (Table 4). Gross motor functioning appeared to be strongly related to reported unmet needs (Table 5). Participants with lower levels of gross motor functioning reported significantly more unmet needs on all domains of the SNAQ, except for family, finances and formal health services (Table 4, Figure 2).

Similarly, participants with a quadriplegia reported significantly more unmet needs on information, finances and formal social services than participants with a hemiplegia.

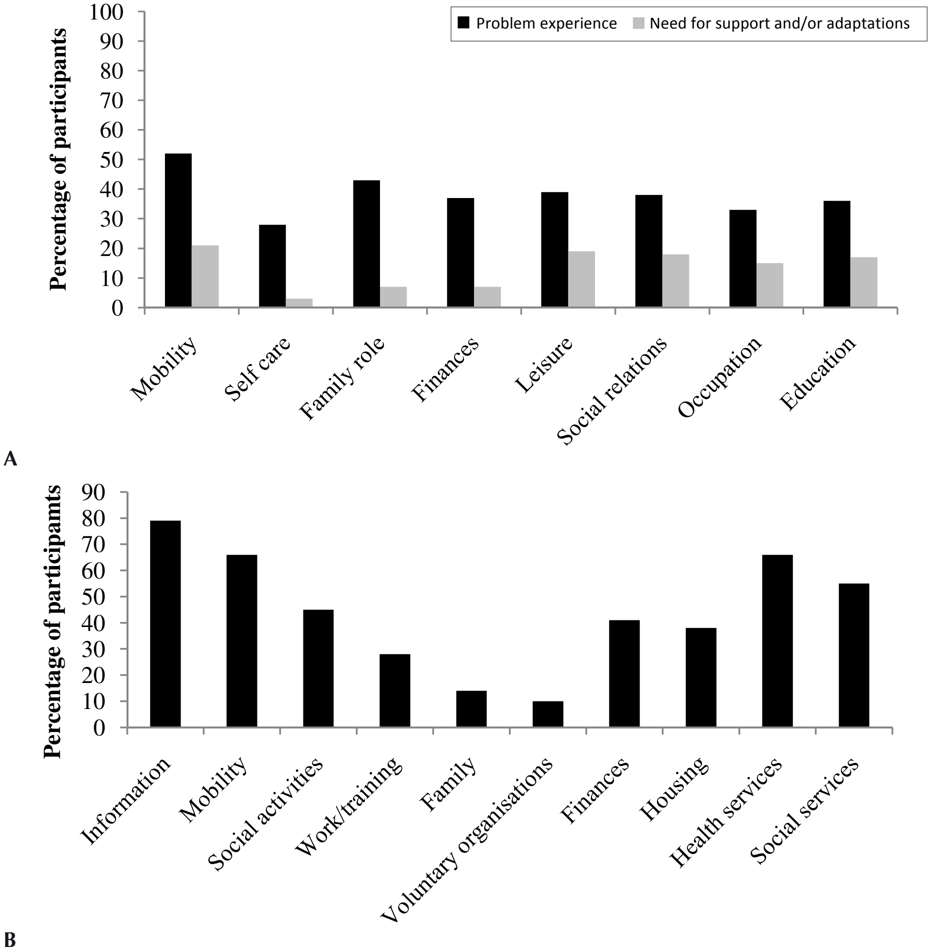

Figure 1. **(a)** Unmet needs as reported on the IPA. **(b)** Unmet needs as reported on the SNAQ.

Finally, participants with lower educational level reported more unmet needs on work or training than those with higher education. No relations were found between unmet needs and age or gender. Figure 2 shows that, although they have less unmet needs than young adults with lower levels of gross motor functioning on some domains, young adults with CP with higher levels of gross motor functioning still indicate several unmet needs.

Health care utilization

Three quarters of the participants (76%) visited a medical specialist, with a rehabilitation physician visited by 52% of the participants (Table 5). Almost three quarters of the participants used allied health care at least once in the past year. Of these, the physical therapist (55%) and social services (32%) were the most frequently visited professionals. Three participants did not make use of any medical care in the past year. One of them did not report unmet needs; the other two participants reported unmet needs on information, mobility and health care. With respect to medical interventions, we found that four participants used some kind of medication for their CP (not for epilepsy). Three participants received a splint in the past year, two underwent

Table 3. Unmet needs mentioned most often on the need domains of the SNAQ.

SNAQ needs domain	**Number of unmet needs (Domain score, 0-100) (Mean (SD))**		**Item mentioned most often *(n)***
Information	34.5	(26.4)	Complications of CP (17)
Mobility	13.2	(13.4)	Changes by other people within the community (11)
Social activities	21.3	(30.8)	Making arrangements for a (adapted) holiday (9)
Work or training	10.4	(20.4)	Help or training to find new work (6)
Family	4.1	(12.4)	Help arranging future care arrangements (2)
Voluntary organizations	10.4	(31.0)	Services from voluntary organization (3)
Finances	21.8	(32.5)	Get benefits (7)
Housing	16.6	(26.2)	Help with maintenance of specialised equipment and wheelchairs (7)
Formal health services	18.1	(21.8)	Getting more physiotherapy (12)
Formal social services	10.3	(12.6)	Counselling for CP-related problems (10)

SNAQ, Southampton Needs Assessment Questionnaire

Table 4. Relations (Spearman) between subject characteristics and number of unmet needs on the SNAQ or treatment by a rehabilitation physician/physical therapist.

	Age	**Gender**	**Level of education**	**Limb distribution**	**GMFCS**
Unmet needs (domain scores)					
Information	-0.36	-0.26	0.02	0.50**	0.48**
Mobility	-0.26	-0.31	0.03	0.28	0.49**
Social activities	-0.08	-0.26	-0.35	0.35	0.62**
Work or training	-0.29	0.14	-0.38*	0.26	0.38*
Family	0.11	0.12	0.34	0.26	0.15
Voluntary organizations	0.06	-0.008	-0.33	0.33	0.47*
Finances	-0.13	-0.098	-0.13	0.55**	0.26
Housing	-0.10	-0.093	0.12	0.35	0.48**
Formal health services	-0.18	-0.160	-0.12	0.11	0.36
Formal social services	0.11	0.060	0.06	0.44*	0.44*
Health care utilization					
Rehabilitation physician	-0.11	-0.03	-0.20	0.48**	0.44*
Physical therapist	0.21	-0.08	-0.18	0.55**	0.55**

* Significant at p≤ .05.
** Significant at p≤ .01.

orthopaedic surgery and one participant received plaster. None of the participants underwent a Botulin toxin A treatment in the past year.

Relations were found between subject characteristics and utilization of medical specialist or allied health care (Table 4, Figure 3). Participants with lower levels of gross motor functioning visited a rehabilitation physician and a physical therapist more than participants with higher levels of gross motor functioning. Similar results were found for the orthopaedist, occupational therapist and speech therapist (Figure 3). Also, more participants with a quadriplegia compared to those with a hemiplegia visited a rehabilitation physician or physical therapist. No relations were found be-

Table 5. Number (%) of young adults with cerebral palsy making use of health care services in the past year.

Health care professional	Number of participants (%)	
Medical specialists	***22***	***(76)***
Rehabilitation physician	15	(52)
Orthopaedist	7	(24)
Urologist	4	(14)
Neurologist	3	(11)
Paediatric physician	0	(0)
Allied health care	***19***	***(68)***
Physical therapist	16	(55)
Social services	9	(32)
Occupational therapist	8	(28)
Psychologist	4	(14)
Speech therapist	2	(7)
Dietician	2	(7)

Missing data on: allied health care, social services and neurologist (n=1)

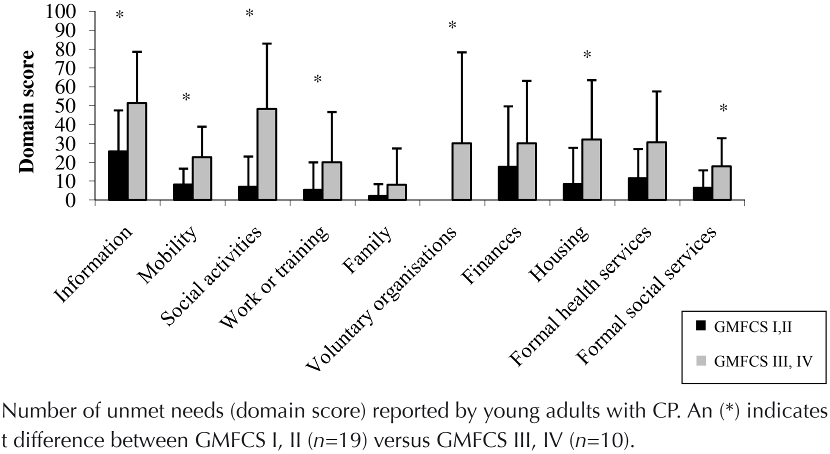

Figure 2. Number of unmet needs (domain score) reported by young adults with CP. An (*) indicates a significant difference between GMFCS I, II (*n*=19) versus GMFCS III, IV (*n*=10).

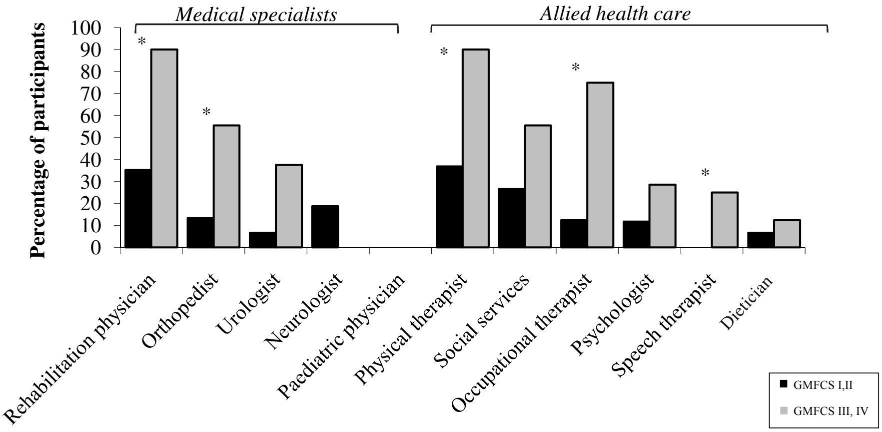

Figure 3. Health care utilization by young adults with CP. An (*) indicates a significant difference between GMFCS I, II (*n*=19) versus GMFCS III, IV (*n*=10).

tween treatment by a rehabilitation physician or physical therapist and age, gender or level of education of the young adults with CP (Table 4).

DISCUSSION

It became apparent in this study, that several unmet needs exist in young adults with CP. The reported unmet needs vary greatly between the participants (both number and type of unmet needs). Overall, the results show that young adults with CP reported unmet needs mostly on information, mobility and health services. Furthermore, the results show that young adults with low gross motor functioning have more unmet needs and a higher utilization of health care than those with higher gross motor functioning. The rehabilitation physician and physical therapist were the most visited health care professionals.

Unmet needs

Our results on the IPA show that being restricted in certain activities does not necessarily lead to an experience of inadequate support/adaptations and a related unmet need. Overall, this study shows that young adults with CP experience unmet needs mostly on information, mobility and health services.

The young adults' need for more information may be explained by the fact that most parents are informed about CP and its consequences when the child is young and is still in treatment with a rehabilitation physician. However, this information might not be forwarded properly to the child as it grows up, leaving the child with several unanswered questions regarding CP and its consequences. Furthermore, questions regarding CP might change, as the needs change when a child reaches adolescence and adulthood.[12,27] Within the domain of social services, the unmet need reported most was counselling for CP-related problems, which is consistent with the need for information. These findings with regard to a need for information are comparable to other studies on children and adolescents with a disability or chronic health condition.[15,28-33]

One of the main consequences of CP is impaired motor functioning. Therefore, problems with mobility are often experienced by people with CP, also in adults.[13,34-36] Problems in mobility may also occur related to environmental factors, such as in the context of the community (i.e. access to public services) and attitudes of other people towards the impaired person (i.e. willingness to help). Our finding that unmet needs on mobility still exist in young adults may imply that the problems with mobility as a result of CP are not (yet) solved in the current health care and environment.

Besides unmet needs regarding information and mobility, many of the young adults with CP reported unmet needs with respect to health care. They felt the need for (more) physiotherapy or would like to visit a medical specialist for a specific problem. This could be the consequence of both changing needs at this age and a discontinuity in health care between paediatric and adult care. In several studies, a discontinuity of care was reported when young adults with physical disabilities entered adulthood and a diminished contact with health care was reported after leaving school.[13-17] Although regular health care utilization has been reported in the present study, it seems that this is not sufficient to fulfil the unmet needs of young adults with CP.

Relations

With respect to the question of which participants reported the most unmet needs and had the highest healthcare utilization, a clear relationship was found with gross motor functioning. We found that participants with lower levels of gross motor functioning and participants with a quadriplegia had more unmet needs, and that they were more likely to be treated by several health care professionals (i.e. rehabilitation physician and physical therapist). Nevertheless, also young adults with higher levels of gross motor functioning or with a hemiplegia still have several unmet needs, while they are less often treated than the participants with lower levels of gross motor functioning or a quadriplegia. Two of the three participants who did not make use of any medical care in the past year also reported unmet needs on several domains.

Limitations

Since no specific instruments were available for measurement of the unmet needs of young adults with CP, we adapted a need-assessment questionnaire (originally developed for stroke patients) and we used part of a generic instrument for participation and autonomy. Both instruments are checklists and this type of questionnaire can tempt the participants to simply say "yes" on the need questions more easily, for example on the question regarding information; this may have resulted in an overestimation of unmet needs. An alternative approach to overcome this problem could be the use of open interviews. In an ongoing study in 87 young adults with CP we use open interviews to establish unmet needs.

Although found in a relatively small sample, we believe that our results can be generalized to other young adults with CP without severe learning disabilities (IQ>70). The young adults were recruited in two rehabilitation centres with regional function with the help of rehabilitation physicians. In Dutch medical practice, consultants in rehabilitation medicine play very much a coordinating role in the integral medical care of CP children.[37] The generalization of our results to other countries has to be studied in more detail. The health care system in each country differs, and unmet needs in other countries may vary greatly due to differences in health care systems and health care legislations.

Implications

Several studies have stressed the importance of adequate care for young adults with physical disabilities during the transition from childhood to adulthood.[6,10,13,14] A lack of coordinated services is often mentioned and multidisciplinary handicapped adult teams are proposed in different studies.[6,26,33,38-40] Specialized young adult teams (YATs) have been formed in the United Kingdom, aimed at smoothing the transition of childhood care to adulthood care. These YATs are multidisciplinary teams developed specifically to facilitate the transition from childhood to adulthood services, which typically include a consultant in rehabilitation medicine, a psychologist, therapists and a social worker.[6,38,40] An evaluation of the cost-effectiveness of YATs showed positive results.[6] In the Netherlands, the formation of specialized rehabilitation teams for young adults with a physical disability is ongoing.[41-43] According to the current study, topics such as information, mobility, work and education, and health care are important aspects that should be incorporated in the treatment of young adults with CP. The results of the current study are incorporated in a Young Adult Team, which is now running in Rijndam Rehabilitation Centre and in the Erasmus Medical Centre in Rotterdam, The Netherlands.[41-43]

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CHAPTER 7 General discussion

GENERAL DISCUSSION

Life expectancy for well-functioning persons with cerebral palsy (CP) is close to that of the unaffected population.[1] Despite these high survival rates, however, consequences of CP in adulthood are often poorly understood.[2] At the same time, as persons with CP enter adulthood, their attendance to rehabilitation care seriously declines.[3,4] From clinical practice and literature, we know that adults with CP often return to rehabilitation care with increasing health problems, such as deterioration of spasticity or fatigue.

In this thesis we aimed to establish modifiable factors at which rehabilitation care in adults with CP may be aimed, in order to have starting points for optimizing rehabilitation care for this group. First, we examined the level of everyday physical activity (PA) and physical fitness and their role in health problems, participation and health-related quality of life (HRQoL) in adults with bilateral CP. Secondly, we assessed the problems that young adults with unilateral or bilateral CP experience themselves in daily life, which may decrease wellbeing at later ages.

In chapter 2 we evaluated the level of everyday PA in 56 ambulatory and non-ambulatory adults (25-45 years) with bilateral spastic CP, using an accelerometry-based Activity Monitor (AM). For 42 ambulatory adults from this original cohort, we determined levels of physical fitness and interrelations between physical fitness, objective and subjective level of everyday PA and fatigue (chapter 3). Also, relations between level of everyday PA, physical fitness and participation and HRQoL were explored in this sample (chapter 4). In the study described in chapter 5, we performed a cross-sectional study in 87 young adults aged 18 to 22 with unilateral or bilateral CP to evaluate the problems experienced by persons with CP themselves. We categorized these problems according to the domains of the ICF. In the final chapter, we used questionnaires to assess the unmet needs and level of health care utilization in 29 adults with CP aged between 18 and 39 with either unilateral or bilateral CP (chapter 6).

MAIN FINDINGS

Level of everyday physical activity and physical fitness

Adults with bilateral spastic CP are at risk for an inactive lifestyle (chapter 2). On average, adults with CP were dynamically active for nearly 2 hours a day (116 minutes). This is significantly less than able-bodied age-mates (157 minutes). Primarily adults with low-level gross motor functioning were at risk for an inactive lifestyle. However, participants with high-level of gross motor functioning, who had comparable levels of everyday PA as able-bodied controls, did also not achieve minimum exercise recommendations for able-bodied persons.[5] Ambulatory adults with bilateral CP had low levels of physical fitness (77% of Dutch reference values[6]) and 50% of the participants experienced fatigue (chapter 3).

An inactive lifestyle in CP was also reported in studies of diplegic children[7] and adolescents.[8-10] With different measurement procedures, including doubly-labeled water, step counts and questionnaires, lower levels of everyday PA were demonstrated in persons with CP compared with able-bodied age-mates. In a recent publication of our department, accelerometry-based levels of everyday PA were reported for several patient groups, among which our sample of adults with bilateral CP, myelomeningocele (MMC), spinal cord injury (SCI) and liver transplantation.[11] Adults with bilateral CP were less physically active than those with e.g. liver transplantation but more physically active than those with MMC and SCI. Furthermore, consistent with the results in our sample on level of gross motor functioning, ambulatory status has been found to be an important determinant of level of PA in persons with MMC[12] and SCI.[13]

The low levels of physical fitness are in agreement with results found in earlier studies in adults[14-15] and in children and adolescents[16-20] with CP. Studies on fatigue in Norway also showed similar results as our study: 30% of participants experienced substantial fatigue, and 17% chronic fatigue, mainly those with bilateral CP.[21-23]

Relations between everyday physical activity, physical fitness and fatigue

Opheim et al. (2009)[23] hypothesized 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. Only in men rather weak relations were present between physical fitness and fatigue. Also, relationships between objective and self-reported PA and fatigue were not present, which is in line with other studies in adults with CP.[22,24] It seems that other factors may play a role in the existence of fatigue in CP. Factors such as behavioral factors should possibly be included in a more psychological-oriented model of fatigue.

The lack of relationships between physical fitness and level of everyday PA was also noteworthy. Although we found a moderate positive relationship between physical fitness and self-reported PA in women, we did not find relationships in men or for the objective measurement of PA. In other patient groups, results on relationships between physical fitness and level of everyday PA are also inconclusive. Some studies reported moderate relationships[11,24] and others reported no relationships.[26,27]

We can speculate on several reasons why we did not find strong interrelations between level of everyday PA, physical fitness and fatigue.

First, other aspects of PA and physical fitness than measured in the studies described in this thesis may be more strongly related to each other or to fatigue. Factors such as the reaction of the body to PA (e.g. energy expenditure, heart rate) and the number of periods of everyday PA of more than 10 minutes at a certain rate of cardiorespiratory strain could have yielded different results in the interrelations between PA, physical fitness and fatigue.

Secondly, possibly the imbalance between physical fitness and level of everyday PA may have had a stronger influence on fatigue than physical fitness or PA itself.

Third, the lack of relations in our study between 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. The strain may not be high enough to induce changes in physical fitness. However, in non-ambulatory persons with MMC, who have low levels of PA, a relationship between level of everyday PA and physical fitness was found.[10] Perhaps a particular threshold is present in level of everyday PA and/or physical fitness below which the two constructs are related.

Furthermore, differences in relationships may be present within our sample depending on walking difficulty: persons with CP who walk with more difficulty have a higher energy expenditure during walking than persons who walk with less difficulty. This may lead to daily training stimuli with regard to physical fitness in those who walk with more difficulty.

Finally, in general it is important to recognize that the level of everyday PA measured in our study is a momentary view of the level of PA during a person's life (not habitual PA), while physical fitness is a capacity which a person builds up throughout his or her life.

Relations between everyday PA, physical fitness, participation and HRQoL

Adjusted for gender and gross motor functioning, objective level of everyday PA was not related to either participation or HRQoL. We did find several relations between physical fitness and the latter parameters (chapter 4). The absence of a relationship between level of everyday PA and participation was contrary to what we expected. In adults with unilateral CP, only participation in sports was associated with level of everyday PA.[28] In other patient groups such as persons with MMC and SCI, the level of everyday PA did play a role in participation.[25,29] Possibly, the variation in level of everyday PA was not large enough in our sample to have strong effects on participation in daily activities and social roles. In addition, everyday PA is a behavior related to the movements that people perform, and the level of everyday PA may not have been high enough to relate to difficulty in participation as assessed with the Life Habits questionnaire.

Adults with lower levels of physical fitness tended to experience more difficulties in daily activities and social roles and had a lower physical HRQoL. Previous studies on relationships between lifestyle and HRQoL in CP used PA as a measure for lifestyle[30,31], yielding similar results as the present study in that no[30] or only weak[31] relationships between PA and HRQoL were found. To our knowledge, no studies are available in adults with CP with regard to relationships between physical fitness and HRQoL. In MMC, Buffart and co-workers[28] showed a similar relationship between physical fitness and participation in young adults. However, physical fitness plays a less important role in participation in patients with SCI.[25]

Experienced problems

At young adult age (18-22 years), persons with CP experienced problems in several areas of daily life, which primarily include mobility, work, self-care, domestic tasks and leisure activities (chapter 5). Problems in these areas were frequently reported and were of high importance for young adults with CP. Persons with low-level gross motor functioning and low-level manual ability appeared to be more prone to experiencing problems in daily life. The aforementioned problem areas are also often described in other studies on young adults and adults with CP.[32-36] So, although much attention is given to some of these problems in pediatric rehabilitation, it appears that these problems remain important at later age.

Adults with CP reported unmet needs mostly on information, mobility and health care (chapter 6). Both number and type of reported unmet needs varied greatly between the participants. Furthermore, adults with low-level gross motor functioning had more unmet needs and a higher utilization of health care than those with high-level gross motor functioning. Nevertheless, those with higher levels of gross motor functioning still had several unmet needs.

Our findings with regard to a need for information on for example the consequences of CP are in line with other studies on children and adolescents with a disability or chronic health condition.[37-39] We explained this need for more information by a possibly inadequate transfer of information previously provided to the parents of a child with CP to the person with CP him- or herself at later age.

Mobility issues are often reported in literature[32,33,40] and could be related directly to the impaired motor functioning in CP (such as walking long distances), but also may occur related to environmental factors, such as access to public services and attitudes of other people towards an impaired person (i.e. willingness to help).

The unmet needs reported with regard to health care services (i.e. getting more physical therapy) might result from both changing needs at adult age and a discontinuity between pediatric and adult health care. Other studies also reported on this discontinuity of health care.[37,41,42]

Summarizing, in our studies on the description of factors at which rehabilitation care in adults with bilateral spastic CP may be aimed, we found several issues. Adults with CP have low levels of everyday PA and physical fitness, of which mainly physical fitness is related to fatigue, participation and HRQoL. Self-reported problems comprise amongst all information, mobility issues and work. Both for PA and physical fitness and for self-experienced problems, persons with low levels of gross motor functioning appear to be at major risk.

METHODOLOGICAL CONSIDERATIONS

Generalizability

In this thesis we described the results of two multi-centre studies performed in the south-western part of the Netherlands. Eligible participants were recruited from several university hospitals and rehabilitation centres throughout this region. These regions covered the most densely populated part of the country. We used a broad approach to include participants, by searching the historical registers of rehabilitation centres and actively approaching the patients' association (BOSK). Response rates were rather low and non-response was mainly due to a lack of up-to-date contact information for eligible participants.

For the three studies, we found no significant differences between responders and non-responders on important characteristics; and reasons for not participating were divers. Main reasons for not participating in the study were overall physical/psychological burden of the research, problems with the duration of the measurement, comorbidities, communication problems and no interest. Given the lack of differences between responders and non-responders and the divers reasons for non-participation, we do not expect that the low response rates have flawed our results.

However, some limitations to the generalizability of our results should be mentioned. First, we did not include adults with unilateral CP in our studies on level of everyday PA, since in our research group prior to the studies described in this thesis, a study was conducted on level of PA and participation in adults with unilateral CP by van der Slot and co-workers.[31] In this previous study, no difference was found in the level of everyday PA between adults with unilateral CP and able-bodied age-mates.

Secondly, several exclusion criteria were formulated. For the studies on PA and physical fitness (chapter 2, 3 and 4), we excluded persons who were fully dependent on electric wheelchair propulsion and who were legally inable. In the studies involving physical fitness, we included only ambulatory adults with CP from the original cohort. Participants were required to perform a cycle ergometer test, which was one of the additional inclusion criteria for these studies. For the studies on experienced problems (chapter 5 and 6), we excluded persons with severe cognitive disabilities, since these studies focused on young adults with CP who have the capacity to become independent participants of our society. This exclusion criterion resulted in a relatively well-functioning cohort of young adults with CP for these studies. We suggest that other studies are needed to develop and evaluate appropriate interventions for persons with CP that are completely bound to an electrical wheelchair or persons with severe intellectual disabilities.

Notwithstanding the abovementioned limitations, we believe that our results are indicative for a broad spectrum of persons with CP. Persons with CP form a heterogeneous patient group with many possible consequences of the condition, so generalization is hard to obtain for the total patient group. However, we included persons with CP from several sources and we measured many aspects of functioning in the participants to obtain a broad picture of consequences of CP regarding lifestyle and health problems. These insights may contribute to the optimization of treatment for adults with CP.

Design of the study

We need to take into account that the studies described in this thesis are cross-sectional and that we cannot infer causality in the relationships that we studied, for example the relationships between level of everyday PA, physical fitness and fatigue. Furthermore, the strength of most relations we reported was moderate. Therefore, results on relations between for example fatigue and physical fitness (chapter 2) and personal characteristics and experienced problems (chapter 6) should be interpreted with caution. In chapter 4 we described the role of level of everyday PA and physical fitness in participation and HRQoL by means of logistic regression analysis, because data on participation and HRQoL were not normally distributed. We dichotomized the outcomes according to cut-off points established in earlier studies (participation) and the mean of a reference population in the United States (HRQoL). In this type of regression analysis information in the outcome measures may be lost and the decision-making process for reference points in dichotomizing scores may somewhat influence the outcomes. However, in this data set, logistic regression was the most appropriate statistical analysis to use, and reported relationships were not very different from relationships found in similar studies in adults with CP.

Outcome measures

Level of everyday physical activity

We measured level of everyday PA with an accelerometry-based AM. Level of everyday PA was measured during a 48-hour period on two randomly selected consecutive weekdays. Although it has been suggested that at least three to five days of measurement may be necessary to characterize habitual PA patterns[43], there is some evidence that a 24-hour measurement with the AM is enough to assess activity levels.[44] Variance between the first and second measurement day was small for our participants, suggesting that two randomly selected consecutive days is sufficient to adequately assess level of everyday PA. Moreover, our aim was to obtain insight in the level of everyday PA as compared with able-bodied persons and not to determine habitual PA.

Also, we may have underestimated level of everyday PA because participants could not swim during measurements. However, able-bodied controls were also not allowed to swim and although swimming is noted in literature as a frequent PA of persons with CP[45,46], in our sample 9% of the participants reported swimming as a frequent exercise. On the other hand, overestimation of level of everyday PA may also have been induced by a selection bias based on the fact that people with interest for sports possibly are more prone to participate in this type of research.

The size and the wires of the AM could also have hampered some activities in daily life. We asked the participants whether they performed their regular activities during the measurement period and when a measurement day was denoted by the participant as being not representative for regular activities, we excluded that day from our analyses. So, overall we considered the influence of the measurement equipment itself to be little.

Physical fitness

We used aerobic capacity expressed as VO_{2peak} in L/min as indicator for physical fitness. Some studies prefer to report on values of VO_{2peak} normalized for body mass (mL/kg/min) or fat-free mass (mL/kgFFM/min). In our regression analyses we used VO_{2peak} in L/min as variable. VO_{2peak} was measured with cycle ergometer tests, which is a non-weight bearing activity and the influence of body mass was expected to be small. Furthermore, there were no excess differences in body weight between the participants measured in our studies. When applicable, we used VO_{2peak} values corrected for gender and level of gross motor functioning. Also, we did additional analyses afterwards with VO2 $_{peak}$ corrected for body mass (mL/kg/min) and found similar results. We did not perform additional analyses with VO_{2peak} corrected for fat-free mass, since the estimation of fat-free mass would be based on skinfold measurements using algorithms from an able-bodied reference population.[47] In children with CP, these algorithms were found to be invalid.[48]

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[6], but values based on maximal cycle ergometer tests were not present for the Dutch situation. However, similar outcomes were found when we compared our results with reference values from Shvartz & Reibold[49], which were measured directly with maximal tests in untrained persons in the United States, Canada, and seven European countries.

Experienced problems

To categorize the experienced problems of young adults with CP in a broad perspective, we linked these problems to the International Classification of Functioning (ICF)[50] in chapter 5. Although categorizing outcomes of a given measurement instrument in another classification system may impose problems and may lead to possible loss of information, we did not experience these problems in our study. The process of linking experienced problems on the COPM to ICF domains was easy and provides a way to further categorize problems that young adults with CP experience in daily life. Describing these problems in terms of the ICF might increase the comparability of studies into the experienced problems of young people with disabilities using different assessment methods.[51,52]

No specific instruments were available to assess unmet needs in adults with CP. Therefore, we adapted a need-assessment questionnaire originally developed for stroke patients. We also used part of a generic measuring instrument for participation and autonomy. Both instruments are checklists and this type of questionnaire can tempt the participants to say “yes” on the need questions more easily; this may have resulted in an overestimation of unmet needs. In the other hand, we have to keep in mind that questionnaires can also induce recall bias and socially desirable answers.

CLINICAL IMPLICATIONS

This thesis on adults with CP combines information on level of everyday PA, physical fitness and their relations to fatigue, participation and HRQoL with information on experienced problems. We believe that insight in both areas is warranted to be able to identify factors that are important with regard to the optimization of rehabilitation care of adults with CP.

In this thesis we hypothesized that level of everyday PA and physical fitness are important factors to consider. To some extent we proved the importance of these factors, both regarding health problems and participation and HRQoL. Health professionals should be aware of the risk of an inactive lifestyle and low levels of fitness with also possible secondary effects on other factors. Results showed that many adults with CP have low levels of everyday PA and physical fitness. In addition, it appears that primarily physical fitness is associated with fatigue, participation and HRQoL.

We do not know whether adults with CP themselves also consider PA and physical fitness to be important. In the studies on experienced problems, participants did not directly mention level of everyday PA and physical fitness as problems. However, possible consequences of low levels of everyday PA and physical fitness where frequently reported, such as problems in mobility (e.g. walking long distances, standing for a long time) and leisure activities (e.g. sport activities). For optimizing treatment of adults with CP a complete picture is important, both incorporating health-related factors that are known to be important based on scientific research and clinical practice, as well as factors indicated by adults with CP themselves. Rehabilitation professionals should be aware that new types of problems may arise when a young adult with CP reaches adulthood, which may warrant another approach or interventions than in pediatric care. It is important to consider patient experiences and priorities in setting treatment goals for adults with CP.

The results of the studies described in this thesis were used in two different clinical initiatives. First, we used the results to develop age-appropriate interventions in specific areas of participation (in addition to regular rehabilitation appointments) for young adults with physical disabilities. Several studies have stressed the importance of coordinated services for young adults with physical disabilities and multidisciplinary young adult teams (YATs) are often proposed.[3,53-56] In the Erasmus Medical Centre and Rijndam Rehabilitation Centre, an outpatient rehabilitation program[57] is offered, which aims to support young adults with physical disabilities in several areas: work[58], household chorus, directing one's own life, pursuing an active lifestyle, sports participation[59], and dating and sexual development.[60,61] The results of the studies described in this thesis have contributed to some interventions in this program. In the Netherlands, several other rehabilitation centers are developing and implementing interventions and tools for YATs within a national network of rehabilitation centers and professionals engaged in improving transition care for young people with a disability.

Second, recently a research initiative called Learn2Move has started in several rehabilitation centers in the Netherlands. In this national research program the effects and mechanisms of age-appropriate interventions for children, adolescents and young adults with CP on mobility-related activity are studied. In Learn2Move 16-24, the Erasmus Medical Center and Rijndam Rehabilitation Center collaborate with VU medical center, Sophia Rehabilitation and Rehabilitation Centre de Hoogstraat to perform a randomized controlled trial (RCT) on the effectiveness of the intervention "Active lifestyle and sports participation in adolescents and young adults with CP aged 16 – 24 years".[62] In this RCT, several outcomes are compared between a group of persons with CP receiving the intervention and a group of persons with CP receiving no intervention. Besides PA and physical fitness, effects on fatigue, participation and HRQoL are evaluated, both on the short- and long-term.

RECOMMENDATIONS FOR FUTURE RESEARCH

Everyday physical activity and physical fitness

Both in the measurement of everyday PA and physical fitness and with regard to the topics of research aimed at these factors, some suggestions can be made.

In our studies we measured level of everyday PA objectively with an accelerometry-based AM. Although the AM we used provides detailed information on level of everyday PA, some limitations are present with regard to the AM. Measurements are restricted to a maximum of 48 hours, the device is rather large and has many wires and bathing or showering is not possible during activity monitoring. Recently, an improved AM has been developed following remodeling projects with the AM used in our studies which makes it more user-friendly and allows more days of measurement.

Furthermore, more information should be gathered about the influence of wearing the AM on wheelchair propulsion in persons with CP. In spinal cord injury, Bussmann and co-workers showed that there was no systematic influence on the amount of daily manual wheelchair propulsion even though low to moderate burden was experienced.[63]

In the study on level of everyday PA (chapter 2) we determined the periods of continuous dynamic activities for at least 5 and 10 minutes per day. To maintain and promote health, able-bodied persons are recommended to engage in 30 minutes of moderate intensity PA each day, which can be divided into 10-minute intervals.[5] Most adults with CP in our study did not achieve this minimum recommended level for the maintenance of health. However, it is uncertain whether guidelines for the general population are appropriate for persons with disabilities such as CP[64]; this should be further studied.

In general, the low levels of physical fitness found in our sample 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 favorable effects on physical fitness in both adults[65] and children[17,19] with CP.

We failed to demonstrate relations between level of everyday PA and physical fitness. This may amongst all 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 heart rate or oxygen uptake during everyday physical activities, could give us more insight in this possible mechanism.

Also, the dose-response relationship between level of everyday PA and physical fitness may be important to consider in future research, since this may yield interesting results considering the relations between PA and physical fitness in terms of for example threshold values. As pointed out in a literature review performed in 2001, it is not possible to conclude whether activity or physical fitness is more important for health. More research with regard to these relations is needed.[66]

In this thesis we excluded persons who were fully dependent on electric wheelchair propulsion and/or legally unable. In a separate study, problems of this subgroup of persons with CP could further be investigated and maybe specific interventions can be developed adjusted to the experienced problems in e.g. PA and physical fitness. These interventions will probably be different from interventions suitable for ambulatory persons with CP or those who use a manual wheelchair. Although the measurement protocol for those who are fully dependent on electric wheelchair propulsion possibly is more complicated than the protocol used in this thesis, it is important to consider this subgroup, since their experienced problems may be even larger than in the current sample.

Experienced problems

Our aim in this thesis was to establish factors at which rehabilitation care in adults with CP may be aimed, in order to have starting points for optimizing rehabilitation care for this group. We consider the opinions of the persons with CP themselves as an important source of information. In our studies on experienced problems we pointed out that therapists and physicians should be aware that new types of problems may arise when a young adult with CP reaches adulthood and will develop an adult lifestyle. This changed perspective may warrant another approach or interventions than in pediatric care. We performed this study in young adults with CP without a particular intervention; it would be interesting to determine whether the same problems and priorities are still present after interventions such as the outpatient treatment rehabilitation program described above.[57] Evaluation of these interventions based on the experienced problems are needed to improve rehabilitation care for adults with CP.

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Summary

SUMMARY

In the introductory **chapter 1** we begin with describing the diagnosis of cerebral palsy (CP) and more specifically possible consequences of CP in adulthood. CP is a frequently occurring condition in childhood and in the Netherlands the average prevalence over 12 birth years (1977-1988) was 1.51 per 1000 inhabitants. In many studies a return to rehabilitation care at adult age is reported for treatment of several health problems. Therefore, it is important to study how rehabilitation care for adults with CP may be optimized. Level of everyday physical activity (PA) and physical fitness are factors at which rehabilitation care could be aimed, since limitations in these factors may contribute to the worsening of health problems when persons with CP grow into adulthood. Research in CP on PA and physical fitness has focused almost entirely on children and adolescents and available studies are described in the introduction. Besides everyday PA and physical fitness, other factors may contribute to the optimization of rehabilitation care for adults with CP. In early adulthood, persons with CP may experience problems, which in turn may lead to exacerbation of health problems at later age. There is an increased awareness about the need for lifespan care of persons with CP. Although the number of lifespan care initiatives is growing, we have limited systematic knowledge of specific problems encountered by young adults with CP during the transition to adulthood. Further knowledge of experienced problems of young adults themselves is of great value in determining relevant topics for interventions. In this thesis we therefore aimed to establish modifiable factors at which rehabilitation care in adults with CP may be aimed, in order to have starting points for optimizing rehabilitation care for this group. Level of everyday PA, physical fitness and self-experienced problems may be possible contributors to an increased level of health problems, which may cause adults with CP to return to rehabilitation care.

Chapter 2 of this thesis describes a cross-sectional study, which we performed to quantify the level of everyday PA in adults with bilateral spastic CP. Both duration of dynamic activities and intensity of activity were lower in adults with CP compared to able-bodied age-mates. We also evaluated the number of periods of continuous dynamic activities for at least 10 minutes per day and only 39% of the participants had one or two of such periods. Following the results of our study, we speculated that these low levels of everyday PA in adults with CP may be explained by e.g. higher energy requirements for daily activities due to reduced muscle mass, inefficient locomotion or limited opportunities for activities such as sports participation. Primarily adults with low-level gross motor functioning were at risk for an inactive lifestyle. However, participants with high-level of gross motor functioning, who had comparable levels of everyday PA as able-bodied controls, did also not achieve

minimum exercise recommendations for able-bodied persons. We speculated that disabled persons may experience increased physical complaints secondary to aging and functional deterioration which may lead to reduced levels of everyday PA. We therefore suggested that also adults with a relatively high-level gross motor functioning could benefit from lifestyles that are more active.

Next, we studied physical fitness and interrelations between physical fitness, level of everyday PA and fatigue. The results of this study are reported in **chapter 3.** Peak oxygen uptake, objective and self-reported levels of PA and level of fatigue were determined in ambulatory adults with bilateral spastic 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 being associated with lower physical fitness levels. Other relationships were not significant. We suggested that the low levels of physical fitness could be attributed to e.g. a lower amount of active muscle mass or a reduction in localized muscle blood flow in the lower extremity muscles. We could not support a theory of deconditioning in which lower levels of everyday PA lead to lower levels of physical fitness, since we did not find consistent relationships between physical fitness and level of everyday PA. Given the absence of strong relationships between fatigue and the other factors in our study, we believe we did not find conclusive evidence for a physical origin of the fatigue experienced in our sample. It seems that other factors may play a role in the existence of fatigue in CP. One could hypothesize that possibly the imbalance between physical fitness and level of everyday PA or other measurements of everyday PA and physical fitness may have a stronger influence on fatigue. In addition, other factors such as behavioral factors should be included in a more psychological-oriented model of fatigue.

In **chapter 4**, we explore physical fitness and level of everyday PA as potential correlates of participation and health-related quality of life (HRQoL) in the same sample as described in chapter 3. We assessed participation in daily activities and social roles with the Life-Habits questionnaire. Physical and mental HRQoL was assessed with the SF-36. Fifty-seven percent of participants experienced difficulties in daily activities and 36% in social roles. Vitality, physical functioning and role limitation scored lowest in physical HRQoL. A higher level of physical fitness correlated to less difficulty in daily activities and social roles and to a better physical HRQoL. The mental aspect of HRQoL was not associated with physical fitness. Variables other than lifestyle factors may play a role in mental well-being, such as self-efficacy, coping strategies, personality, educational level or social support. We also did not find evidence for an association between level of everyday PA and participation or HRQoL; this was contrary to what we expected. We expected the level of everyday

PA to be correlated primarily to participation in daily activities. It is possible that the variation in level of everyday PA was not large enough to have strong effects on participation in daily activities and social roles. In addition, everyday PA is a behavior related to the movements that people perform, and the level of everyday PA may not have been high enough to relate to difficulty in participation as assessed with the Life-Habits questionnaire.

We describe problems experienced by young adults with CP (18-22 years) in a broad sense by using semi-structured interviews in **chapter 5**. In this study, we used the International Classification of Functioning to categorize the experienced problems. Approximately 70% of participants experienced problems in daily life, addressing the areas of self-care, productivity and leisure activities. Persons with low-level level gross motor functioning and low-level manual ability appeared to be more prone to experiencing problems in daily life. Problems were most prevalent in recreation and leisure, preparing meals, housework and dressing. Problems in functional mobility, (un) paid work, and socialization were considered as most important (represented by the highest mean importance score). When growing into adulthood, young adults with disabilities have to learn a range of life skills which enable them to participate in adult life, such as finding a vocation, transport to work and doing housework. The current study showed that young adults with CP rated problems in participation as very important. Although topics such as mobility and self-care are frequently addressed in pediatric rehabilitation interventions, problems in these areas might still exist during (young) adulthood. Furthermore, problems with regard to work and domestic life may appear when the person with CP grows older, which can be indicated as growing into a deficit. Therapists and physicians should be aware that new types of problems may arise when a young adult with CP reaches adult age, which may warrant another approach or interventions than in pediatric care. It is important to consider patient experiences and priorities in setting treatment goals for young adults with CP.

In **chapter 6** we aim to obtain insight in the unmet needs and health care utilization of young adults with CP (aged 18-39). Young adults with CP reported unmet needs mostly on information, mobility and health care. The widely reported need for more information could be explained by the fact that most parents are informed about CP and its consequences when the child is young and is still in treatment with a rehabilitation physician. However, this information might not be forwarded properly to the child as it grows up, leaving the child with several unanswered questions regarding CP and its consequences. Participants with lower levels of gross motor functioning were found to have more unmet needs and visited various health care professionals more often than young adults with higher levels of gross motor functioning. However, participants with higher levels of gross motor functioning still reported several unmet needs, while they are less often treated in medical care than those with lower levels of gross motor functioning. As important implication of the results of this study the availability of coordinated services through specialized young adult teams is discussed to facilitate the transition from childhood to adulthood services.

Finally, **chapter 7** of the thesis describes the main findings of the thesis and discusses the methodological considerations (both strengths and limitations) with regard to the studies described in this thesis. Generalizability, the design of the studies and important outcome measures are evaluated. This is followed by the clinical implications of our studies and recommendations for future research with regard to PA, physical fitness and experienced problems.

Samenvatting (summary in Dutch)

SAMENVATTING

In het inleidende hoofdstuk (**hoofdstuk 1**) beschrijven we de diagnose cerebrale parese (CP) en meer specifiek de mogelijke consequenties van CP op volwassen leeftijd. CP is een vaak voorkomende aandoening. In Nederland is de gemiddelde prevalentie 1.51 per 1000 inwoners, gerekend over 12 geboortejaren (1977-1988). Veel studies laten zien dat volwassenen met CP terugkeren in de revalidatiezorg als gevolg van diverse gezondheidsproblemen. Het is dus belangrijk om te onderzoeken hoe de revalidatiezorg voor volwassenen met CP zoveel mogelijk geoptimaliseerd kan worden en welke factoren hiertoe mogelijk bijdragen. Beperkingen in levensstijl-gerelateerde factoren zoals het niveau van dagelijkse lichamelijke activiteit en fysieke fitheid dragen mogelijk bij aan een verslechtering van de gezondheid. De beschreven studies in de introductie van dit proefschrift laten echter zien dat bijna al het onderzoek in mensen met CP, met betrekking tot lichamelijke activiteit en fysieke fitheid, zich richt op kinderen en adolescenten. Naast het niveau van dagelijkse lichamelijke activiteit en fysieke fitheid kunnen ook andere factoren bijdragen aan optimalisatie van de revalidatiezorg voor volwassenen met CP. Jongvolwassenen met CP ervaren mogelijk specifieke problemen die kunnen leiden tot een verslechtering van de gezondheid op latere leeftijd. Zorgverleners zijn zich steeds meer bewust van het belang van ‘lifespan care’ voor mensen met CP. Hoewel het aantal initiatieven hierin toeneemt, ontbreekt systematische kennis van de specifieke problemen waar jongvolwassenen met CP tijdens de transitie van kind naar volwassene tegenaan lopen. Meer kennis van de problemen zoals jongvolwassenen die zelf ervaren is van grote waarde voor het bepalen van relevante interventies. In dit proefschrift hebben we ons gericht op het vaststellen van modificeerbare factoren waarop de revalidatiezorg voor volwassenen met CP gericht kan worden, om zodoende startpunten te hebben om de revalidatiezorg voor deze groep te optimaliseren. Hierbij richten we ons op factoren die mogelijk bijdragen aan een toename van het aantal of de ernst van de gezondheidsproblemen, hetgeen er weer toe kan leiden dat volwassenen met CP terugkeren in de revalidatiezorg.

Hoofdstuk 2 van dit proefschrift beschrijft een cross-sectionele studie die is uitgevoerd om het niveau van dagelijkse lichamelijke activiteit bij volwassenen met bilaterale spastische CP vast te stellen. Zowel de duur van dynamische activiteiten als de intensiteit van de activiteiten was lager bij volwassenen met CP dan bij leeftijdsgenoten zonder lichamelijke beperking. We hebben tevens het aantal perioden van continue dynamische activiteit gedurende ten minste 10 minuten per dag bekeken. Slechts 39% van de deelnemers had één of twee van zulke perioden. Het lage niveau van dagelijkse lichamelijke activiteit bij volwassenen met CP wordt mogelijk verklaard door onder meer een hoger energieverbruik bij het uitvoeren van dagelijkse activiteiten als gevolg van een lagere spiermassa, inefficiënte voortbeweging of beperkte mogelijkheden voor activiteiten zoals sport. Vooral volwassenen met een laag niveau van grofmotorisch functioneren hadden een verhoogd risico op een inactieve levensstijl. Deelnemers met een hoog niveau van grofmotorisch functioneren hadden vergelijkbare niveaus van dagelijkse lichamelijke activiteit als leeftijdsgenoten zonder een lichamelijke beperking, maar ook zij haalden niet de minimale inspanningsaanbevelingen voor personen zonder een lichamelijke beperking. Ongeacht het niveau van grofmotorisch functioneren, ervaren mensen met een beperking mogelijk een toename in fysieke klachten secundair aan het ouder worden en hun functionele achteruitgang. Deze toename kan mogelijk leiden tot lagere niveaus van dagelijkse lichamelijke activiteit. Daarom menen we dat ook volwassenen met een relatief hoog niveau van grofmotorisch functioneren kunnen profiteren van een meer actieve levensstijl.

Vervolgens hebben we gekeken naar fysieke fitheid en de onderlinge relaties tussen fysieke fitheid, het niveau van dagelijkse lichamelijke activiteit en vermoeidheid. De resultaten van deze studie zijn beschreven in **hoofdstuk 3**. De maximale zuurstofopname (als maat voor de fysieke fitheid), objectieve en zelf-gerapporteerde niveaus van lichamelijke activiteit en het niveau van vermoeidheid werden bepaald bij ambulante volwassenen met bilaterale spastische CP. Deelnemers hadden lagere niveaus van fysieke fitheid, waren minder lichamelijk actief dan leeftijdsgenoten zonder een lichamelijke beperking en de helft van de deelnemers ervoer vermoeidheid. Bij vrouwen was een lage fysieke fitheid gerelateerd aan lagere zelf-gerapporteerde fysieke activiteit en bij mannen was een lage fysieke fitheid gerelateerd aan een hogere vermoeidheid. Andere relaties waren niet significant. Mogelijk kunnen de lagere niveaus van fysieke fitheid worden toegeschreven aan bijvoorbeeld een lagere hoeveelheid actieve spiermassa of een reductie in de bloedtoevoer naar bepaalde beenspieren. We hebben geen ondersteuning kunnen vinden voor een theorie van deconditionering waarin lagere niveaus van dagelijkse lichamelijke activiteit zouden leiden tot lagere niveaus van fysieke fitheid, aangezien er geen consistente relaties waren tussen fysieke fitheid en het niveau van dagelijkse lichamelijke activiteit. Gezien het ontbreken van sterke relaties tussen vermoeidheid en de andere factoren in onze studie, hebben we geen overtuigend bewijs kunnen vinden voor een fysieke oorsprong van de ervaren vermoeidheid van onze deelnemers. Het lijkt dat andere factoren mogelijk een rol spelen in het bestaan van vermoeidheid bij mensen met CP. Mogelijk hebben de disbalans tussen fysieke fitheid en het niveau van dagelijkse lichamelijke activiteit of andere maten van lichamelijke activiteit en fysieke fitheid een sterkere invloed op vermoeidheid. Voorts zouden andere factoren zoals gedragsfactoren geïncludeerd moeten worden in een meer psychologisch georiënteerd model van vermoeidheid.

In **hoofdstuk 4** onderzoeken we de relaties tussen fysieke fitheid en het niveau van dagelijkse lichamelijke activiteit enerzijds en participatie en gezondheidsgerelateerde kwaliteit van leven anderzijds in dezelfde populatie als beschreven in hoofdstuk 3. We onderzochten participatie in dagelijkse activiteiten en sociale rollen met behulp van de Life-Habits vragenlijst. Fysieke en mentale kwaliteit van leven werd vastgesteld met de SF-36. 57% van de deelnemers ervoer moeite met het uitvoeren van dagelijkse activiteiten en 36% met het uitvoeren van sociale rollen. Van de fysieke kwaliteit van leven domeinen, scoorden vitaliteit, fysiek functioneren en rolbeperking het laagst. Deelnemers met een hoger niveau van fysieke fitheid hadden minder moeite met het uitvoeren van dagelijkse activiteiten en sociale rollen en hadden een hogere fysieke kwaliteit van leven. Het mentale aspect van kwaliteit van leven was niet geassocieerd met fysieke fitheid. In plaats van eerdergenoemde levensstijl-gerelateerde factoren spelen mogelijk andere factoren een grotere rol in het mentale welbevinden zoals ‘self-efficacy’, copingstijlen, persoonlijkheid, opleidingsniveau of sociale steun. We hebben evenmin bewijs gevonden voor een associatie tussen het niveau van dagelijkse lichamelijke activiteit en participatie of gezondheidsgerelateerde kwaliteit van leven; dit was tegengesteld aan onze verwachtingen. We hadden verwacht dat het niveau van dagelijkse lichamelijke activiteit vooral gerelateerd zou zijn aan participatie in dagelijkse activiteiten. Mogelijk was de variatie in het niveau van dagelijkse lichamelijke activiteit niet groot genoeg om een sterk effect te hebben op participatie in dagelijkse activiteiten en sociale rollen. Verder is dagelijkse lichamelijke activiteit een gedragskenmerk gerelateerd aan de bewegingen die mensen maken. Het niveau van dagelijkse lichamelijke activiteit is mogelijk niet hoog genoeg geweest om te relateren aan de problemen in participatie zoals deze vastgesteld worden met de Life-Habits vragenlijst.

We beschrijven problemen ervaren door jongvolwassenen met CP (18-22) in brede zin door het gebruik van semi-gestructureerde interviews in **hoofdstuk 5**. In dit onderzoek gebruikten we de *International Classification of Functioning, Disability and Health* om de ervaren problemen verder te categoriseren. Bijna 70% van de deelnemers ervoer problemen in het dagelijks leven, verdeeld over de categorieën zelfverzorging, productiviteit en vrijetijdsbesteding. Personen met een laag niveau van grofmotorisch functioneren en een laag niveau van manuele vaardigheden bleken vaker problemen in het dagelijks leven te ervaren. Problemen kwamen het meest voor in recreatie en vrije tijd, eten klaarmaken, huishoudelijke activiteiten en aankleden. Problemen in functionele mobiliteit, (on)betaald werk en socialisatie werden gezien als het meest belangrijk (gerepresenteerd door de hoogste gemiddelde belangrijkheidsscore). Bij het volwassen worden moeten jongvolwassenen met beperkingen een hele range van ‘life skills’ leren die hen in staat stellen om deel te nemen aan het volwassen leven, zoals het vinden van werk, transport naar het werk en het maken van huiswerk. De huidige studie heeft laten zien dat jongvolwassenen met CP problemen in participatie als zeer belangrijk ervaren. Hoewel onderwerpen als mobiliteit en zelfverzorging vaak aan de orde komen bij interventies in de kinderrevalidatie, bestaan er hier mogelijk nog steeds problemen mee op (jong)volwassen leeftijd. Verder komen er mogelijk problemen met betrekking tot werk en huishoudelijke taken naar voren als de persoon met CP ouder wordt, iets wat kan worden aangegeven met 'growing into a deficit'. Therapeuten en artsen moeten zich bewust zijn van het feit dat nieuwe problemen naar voren kunnen komen wanneer een jongere met CP de volwassen leeftijd bereikt, die mogelijk een andere benadering en andere interventies vragen dan in de kinderrevalidatie. Het is daarom belangrijk om patiëntervaringen en prioriteiten in acht te nemen bij het stellen van behandeldoelen voor jongvolwassenen met CP.

De studie in **hoofdstuk 6** heeft als doel inzicht te krijgen in de behoeften ('unmet needs') en het gebruik van de gezondheidszorg door (jong)volwassenen met CP (18-39 jaar). (Jong)volwassenen met CP gaven vooral behoeften aan met betrekking tot informatie, mobiliteit en gezondheidszorg. De veelal gerapporteerde behoefte aan meer informatie zou kunnen worden verklaard uit het feit dat de meeste ouders worden geïnformeerd over CP als het kind nog jong is en nog steeds onder behandeling is van een revalidatiearts. Deze informatie wordt echter wellicht niet duidelijk doorgegeven aan het kind als hij/zij groter wordt, waardoor het kind blijft zitten met verschillende onbeantwoorde vragen over CP en de consequenties ervan. Deelnemers met lagere niveaus van grofmotorisch functioneren hadden meer behoeften en maakten meer gebruik van diverse zorgverleners dan degenen met hogere niveaus van grofmotorisch functioneren. Echter, degenen met hogere niveaus van grofmotorisch functioneren rapporteerden nog steeds diverse behoeften, terwijl zij minder vaak worden behandeld in de gezondheidszorg. Als belangrijke implicatie van de resultaten van deze studie wordt de beschikbaarheid van gecoördineerde zorgverlening door gespecialiseerde 'young adult teams' besproken om de transitie van de kinderzorg naar de volwassenenzorg te faciliteren.

Tenslotte beschrijft **hoofdstuk 7** van dit proefschrift de belangrijkste bevindingen en bespreken we de methodologische aspecten (zowel de sterke kanten als de beperkingen) met betrekking tot de studies beschreven in dit proefschrift. De generaliseerbaarheid, de onderzoeksopzet van de studies en belangrijke uitkomstmaten worden geëvalueerd. Dit wordt gevolgd door de klinische implicaties van onze studies en aanbevelingen voor toekomstig onderzoek met betrekking tot lichamelijke activiteit, fysieke fitheid en ervaren problemen.

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Beste Henk, daar sta je weer bovenaan in een dankwoord. Waar 99% van de mensen (niet wetenschappelijk onderbouwd) bij een proefschrift als eerste het dankwoord leest, ben jij waarschijnlijk die ene persoon van de 100 die het niet leest. Neemt niet weg dat het voor mij belangrijk is om je te benoemen. Op het moment dat ik je als promotor het meest nodig had, toonde je begrip voor mijn situatie van een in mijn ogen onverenigbare combinatie van een drukke nieuwe baan, een openstaande promotie en een jong gezin. Door jouw begrip en steun werd ik gemotiveerd om door te gaan en hier is dan het resultaat. Uiteindelijk kom ook ik dus hopelijk in het gouden lijstje!

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Curriculum Vitae

CURRICULUM VITAE

Channah Nieuwenhuijsen was born in Spijkenisse on the 8th of March 1982. After finishing her atheneum at OSG De Ring van Putten in Spijkenisse in 2000, she started studying Human Movement Sciences at the Vrije Universiteit in Amsterdam. She completed her study with a master thesis on Behavioral problems in pre-term infants. Furthermore, she did her research internship at the department of Rehabilitation Medicine of the Erasmus Medical Center (Erasmus MC) under supervision of dr. H.J.G. van den Berg-Emons, studying level of everyday PA, fatigue and health-related quality of life in liver transplant recipients. She also obtained her degree as a HBO (Hoger Beroeps Onderwijs) qualified teacher. In 2004, she finished the Master of Science education with a major in Rehabilitation Medicine cum laude.

After her study, she started as a junior researcher at the department of Rehabilitation Medicine of Erasmus MC, in the research project 'CP Transition Study South West Netherlands'. As part of the national network PERRIN (Pediatric Rehabilitation Research in the Netherlands) she performed several interviews with adolescents and young adults with Cerebral Palsy. From 2005 she started as a PhD-student in the research project 'Everyday physical activity and fitness in adults with bilateral spastic cerebral palsy'. The results from both studies led to this PhD-thesis.

Currently, she is working as a quality advisor at the Ruwaard van Putten Hospital in Spijkenisse. Channah lives together with Arnold Hamerslag and their two children Kyana Isa (2008) and Sten Luukas (2010).

List of publications

LIST OF PUBLICATIONS

Nieuwenhuijsen C, van der Slot WMA, van den Berg-Emons, Terburg M, Bergen MP, Stam HJ, Roebroeck ME and the Transition Research Group South-West Netherlands. The role of physical fitness and activity in participation and health-related quality of life in adults with cerebral palsy. **Submitted.**

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Van den Berg-Emons HJG, Kazemier G, van Ginneken BTJ, Nieuwenhuijsen C, Tilanus HW, Stam HJ. Fatigue, level of everyday physical activity and quality of life after liver transplantation. **Journal of Rehabilitation Medicine 2006; 38: 124-129.**

PhD Portfolio
Summary

PHD PORTFOLIO SUMMARY

SUMMARY OF PHD TRAINING AND TEACHING ACTIVITIES

Name PhD student: Channah Nieuwenhuijsen
PhD period: 2004-2009
Erasmus MC Department: Rehabilitation Medicine
Promotor: Prof. H.J. Stam
Research School: None
Supervisors: Dr. H.J.G. van den Berg-Emons, Dr. M.E. Roebroeck

	Year	Workload
General academic skills		
- ‘Onderzoeksmethoden voor het bewegingsapparaat’. Research Institute MUSC, Rotterdam.	2007	32 hours
- Biomedical English Writing and Communication, Rotterdam.	2007	75 hours
Research skills		
- Statistical Course Lineaire regressie en variantie-analyse, Amsterdam	2007	30 hours
In-depth courses		
- ‘Cliëntgericht werken en het gebruik van de COPM’. Pro Education, Erasmus MC, Rotterdam.	2004	59 hours
- Follow-up course ‘Cliëntgericht werken en het gebruik van de COPM’. Pro Education, Erasmus MC, Rotterdam.	2006	8 hours
- ‘Volwassenen met CP’. ITON, Haarlem.	2007	92 hours
Research Group		
- Research meetings, dept. of Rehabilitation Medicine, Rotterdam	2004-2009	160 hours
Presentations		
- Oral presentation ‘Zorgbehoeften en zorggebruik bij (jong) volwassenen bij cerebrale parese’ at the research meeting, dept. of Rehabilitation Medicine, Rotterdam.	2005	12 hours
- Poster presentation ‘Unmet needs and healthcare utilization in young adults with cerebral palsy’ at VRA symposium, Arnhem.	2005	24 hours
- Poster presentation ‘Unmet needs and healthcare utilization in young adults with cerebral palsy’ at the 17th Annual Conference of the European Academy for Childhood Disability, Monaco.	2005	8 hours
- Oral presentation ‘Zorgbehoeften en zorggebruik: hoe meten we dat?’ at research meeting PERRIN, Utrecht.	2005	6 hours
- Oral presentation ‘Zorgbehoeften en zorggebruik van jongeren en jongvolwassenen met cerebrale parese’ at research meeting PERRIN, Utrecht.	2005	10 hours
- Oral presentation ‘Lost in Transition? Ervaren problemen en zorggebruik van jongvolwassenen met cerebrale parese’ at PERRIN symposium, Utrecht.	2006	16 hours
- Oral presentation ‘Dagelijks functioneren en fitheid van volwassenen met een spastische cerebrale parese: eerste resultaten’ at regionale refereeravond regio Utrecht, Utrecht.	2006	16 hours

	Year	Workload
- Poster presentation 'Everyday physical activity in adults with bilateral spastic cerebral palsy' at 19th Annual Conference of the European Academy of Childhood Disability, Groningen.	2007	24 hours
- Oral presentation 'Adults with bilateral spastic cerebral palsy are at risk for an inactive lifestyle' at the 16th European Congress of Physical and Rehabilitation Medicine 'From cell to society', Brugge, Belgium.	2008	12 hours
- Poster presentation 'Adults with bilateral spastic cerebral palsy are at risk for an inactive lifestyle' at the 62nd Annual meeting of the American Academy for Cerebral Palsy and Developmental Medicine, Atlanta, USA.	2008	8 hours
- Poster presentation 'Experienced problems of young adults with cerebral palsy: targets for rehabilitation care?' at the 62nd Annual meeting of the American Academy for Cerebral Palsy and Developmental Medicine, Atlanta, USA.	2008	8 hours
- Poster presentation 'Everyday physical activity in adults with bilateral spastic cerebral palsy' at the International Conference on Ambulatory Monitoring of Physical Activity and Movement (ICAMPAM), Rotterdam.	2008	8 hours
- Oral presentation 'Inactive lifestyle in adults with bilateral spastic cerebral palsy' at 4th International State-of-the-Art Congress 'Rehabilitation: Mobility, Exercise & Sports', Amsterdam.	2009	24 hours
- Oral presentation 'Physical fitness and relationships with everyday physical activity and fatigue in ambulatory adults with bilateral spastic cerebral palsy' at the research meeting, dept. of Rehabilitation Medicine, Rotterdam.	2009	24 hours
- Oral presentation 'Adults with bilateral spastic cerebral palsy are at risk for an inactive lifestyle' at the Symposium of the International Cerebral Palsy Society 'A Global Status Quo on Cerebral Palsy, with a View to the Future', Utrecht.	2009	16 hours
- Oral presentation 'Low health-related physical fitness in adults with bilateral spastic cerebral palsy' at the 63rd Annual meeting of the American Academy for Cerebral Palsy and Developmental Medicine, Phoenix, USA.	2009	2 hours
International conferences		
- 17th Annual Conference of the European Academy for Childhood Disability, Monaco.	2005	32 hours
- 19th Annual Conference of the European Academy for Childhood Disability, Groningen.	2007	24 hours
- 62nd Annual meeting of the American Academy for Cerebral Palsy and Developmental Medicine, Atlanta, USA.	2008	40 hours
- 4th International State-of-the-Art Congress 'Rehabilitation: Mobility, Exercise & Sports', Amsterdam.	2009	24 hours
- Symposium of the International Cerebral Palsy Society 'A Global Status Quo on Cerebral Palsy, with a View to the Future', Utrecht.	2009	8 hours

	Year	Workload
Seminars and workshops		
- Symposium Pediatric Rehabilitation Research in the Netherlands (PERRIN) 'CP: Hoe leef je er mee? Van baby tot volwassene'. Utrecht.	2004	8 hours
- Meeting for participants CP Transition Research, Rotterdam.	2005	6 hours
- Symposium VRA, Arnhem.	2005	8 hours
- Symposium 'Onderzoek kinderrevalidatie in beweging', Amsterdam.	2005	8 hours
- Regionale refereeravond regio Utrecht, Utrecht.	2006	4 hours
- Symposium Pediatric Rehabilitation Research in the Netherlands (PERRIN) 'Kinderen en jongvolwassenen met CP: hun ontwikkeling, onze zorg'. Utrecht.	2006	8 hours
- Symposium 'Spasticiteit bij kinderen en volwassenen met een verstandelijke beperking: hoe te beoordelen en wat eraan te doen?' Rotterdam.	2006	8 hours
- MUSC retraite 'Samenwerken in beweging IV', Rotterdam	2006	8 hours
- Symposium 'Body@work: werken aan bewegen en gezondheid'. Amsterdam.	2007	8 hours
- Symposium '(Hand)cycling Revalidatie en Sport van Werkgroep VRA bewegen en sport', Zuidlaren.	2007	8 hours
Didactic skills		
-		
Other		
- Organizing meeting for participants CP Transition Research, Rotterdam.	2005	16 hours
- Member of organizing committee for symposium Pediatric Rehabilitation Research in the Netherlands (PERRIN) 'Kinderen en jongvolwassenen met CP: hun ontwikkeling, onze zorg'. Utrecht.	2006	16 hours
- Translating the Manual Ability Classification System in Dutch.	2005	16 hours
Teaching activities		
Lecturing		
-		
Supervising practicals and excursions		
-		
Supervising Master's theses		
- Supervising of 2 students Human Movement Sciences (thesis).	2007	32 hours
Other		
- Supervising of a MD in residency.	2006/2007	80 hours
- Supervising a student in Movement Technology.	2008	30 hours
- Assisting co-workers with poster presentations.	2005-2009	12 hours

Total 1080 hours