

ORIGINAL REPORT

MOTOR IMPAIRMENTS AND ACTIVITY LIMITATIONS IN CHILDREN WITH SPASTIC CEREBRAL PALSY: A DUTCH POPULATION-BASED STUDY

Marc Wichers, MD¹, Sander Hilberink, MSc¹, Marij E. Roebroek, PhD¹, Onno van Nieuwenhuizen, MD, PhD² and Henk J. Stam, MD, PhD¹

From the ¹Department of Rehabilitation Medicine, Erasmus MC – University Medical Centre Rotterdam, Rotterdam and ²Department of Child Neurology, University Medical Centre Utrecht, Utrecht, The Netherlands

Objective: To determine the prevalence of motor impairments and activity limitations and their inter-relationships in Dutch children with spastic cerebral palsy.

Patients and methods: In a population-based survey 119 children, age range 6–19 years, with spastic cerebral palsy were examined. Anthropometry, muscle tone, abnormal posture, joint range of motion, major orthopaedic impairments and gross motor functioning and manual ability were assessed or classified, in addition to limitations in mobility and self-care activities. Spearman's correlation coefficients, bivariate *post hoc* analyses and univariate and multivariate logistic regression analyses were used.

Results: Children with spastic cerebral palsy had a lower body height and weight compared with typically developing peers. Forty percent had no range of motion deficits. Hip dislocations were rarely encountered. Motor impairments were associated with gross motor functioning and manual ability levels. Close to sixty-five percent walked independently. Children with diplegia and tetraplegia differed in activity limitations. Motor impairments and limitations in mobility and self-care activities were only modestly related in multivariate analyses.

Conclusion: Distribution of cerebral palsy-related characteristics is consistent with that found in representative studies of other countries. The distinction between diplegia and tetraplegia is relevant from an activity point of view. The child's activity limitations are not a mirror of the motor impairments, which suggests multifactorial influences. An activity-oriented rehabilitation approach goes beyond treating specific impairments.

Key words: cerebral palsy, motor impairments, activity limitations, child, prevalence.

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Correspondence address: M. J. Wichers, Department of Rehabilitation Medicine, Erasmus MC, University Medical Centre Rotterdam, PO Box 2040, NL-3000 CA Rotterdam, The Netherlands. E-mail: mjwichers@gmail.com

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within the group of children with CP and into the relationship between specific impairments and activity limitations may be helpful in directing rehabilitation goals. Quantitative data on CP-related motor impairments and activity limitations can identify phenomena appropriate for longitudinal study, thus promoting adequate planning for both research and health services.

Several population-based CP studies in other European countries have explored prevalence and inter-relationships of clinical features, motor impairments, activities and described appropriate methods of classification (2–7). The inter-relationship of motor impairments and activity limitations is not always straightforward. One issue raised is whether the distinction between leg-dominated and 4-limb dominated spastic CP is relevant in the light of activity limitations or whether describing a Gross Motor Functioning Classification System (GMFCS) level is sufficient to describe the child in this respect (8).

In the Netherlands prevalence data on motor impairments and activity limitations in children with CP has not been available until now. This paper presents representative Dutch data on these issues. We focus on spastic CP, as this form is by far the largest sub-group of CP (1, 9).

According to the World Health Organization's (WHO) International Classification of Functioning, Disability and Health (ICF), impairments are described as significant deviations or loss of body function or body structure (10). In CP, dysfunction of muscle control prevails, which can lead to spasticity or shifting muscle tone, to associated pathological postures, and to decreased range of joint motions.

Activity limitations refer to difficulties in executing tasks or actions (10). The current study focused on limitations in mobility, addressing walking, lifting and arm/hand use, and in self-care activities.

The objectives of this paper are to provide prevalence data on neuromusculoskeletal impairments, (i.e. "motor impairments") and activity limitations in the Dutch population of children with spastic CP and to gain insight into the relationship between impairment and activity limitation.

INTRODUCTION

Children with cerebral palsy (CP) present a variety of clinical presentations and a range of motor impairments and activity limitations (1). Insight into the distribution of these elements

METHODS

Subjects

The present study is part of a cross-sectional population-based survey. Previous publications from this study addressed prevalence and clinical

cal characteristics of CP in the Netherlands (9, 11). CP was defined as a disorder of movement and/or posture caused by a non-progressive brain lesion with an onset no later than one year after birth (12). Obligatory neuromotor disorders (spasticity, dyskinesia or ataxia) were present in all patients. Patients were included if they had: (i) a diagnosis of "cerebral palsy" recorded in their patient files; (ii) date of birth between 1 January 1977 and 31 December 1988; and (iii) parents living, at the time the study was conducted, in Gelderland, a region in the east-central part of the Netherlands. In the present study, we concentrated on children with spastic CP (over 90% of the total group). Children with ataxic or dyskinetic CP were excluded; these non-spastic sub-groups in the cohort each comprised only 4 children. Hence, we present the results of a representative group of 119 children with spastic CP. Informed consent was obtained from the parents of each participant. The study was approved by the ethics committees of the university medical centre and collaborating institutions.

Data collection

Data collection was carried out according to the study protocol of previous German and Swedish studies on the epidemiology of CP (13–16). The protocol and classification criteria were discussed and trained (in advance) together with the authors of these studies.

An experienced child rehabilitation physician visited each child and his or her parents. A structured interview with the parents or caretakers and a physical examination of the child were performed. The interview covered the child's CP-related and general medical history, current and past treatment, adaptations, milestones in development, present performance of activities of daily living, communication, behaviour, school career, and family situation. In the examination, basic characteristics of the child were recorded, e.g. sex, age, body height and weight recorded with a tape measure and household scales, respectively. Body mass index (BMI) was calculated. Intellectual functioning was classified in 3 major levels: (i) normal; (ii) learning disability; or (iii) mental retardation, according to the German-Swedish distinctions (15). The limb distribution of a child's spastic CP was classified as unilateral spastic CP (hemiplegia) or bilateral spastic CP (BSCP). BSCP was subdivided in the leg-dominated form diplegia, or the 4-limb dominated form tetraplegia (17). In leg-dominated spastic CP or diplegia the arms still can be involved and often are, but to a lesser degree than the legs. Gross motor functioning and manual ability were classified according to the GMFCS (18) and the Manual Ability Classification System (MACS) (19), respectively.

Examination of the lower extremities was performed with the child in supine position and for trunk and upper extremities in sitting position. Using manual passive or (assisted active) flexion/extension of the entire extremities in the major joints, the physician judged muscle tone as elevated (1) or not (0) in each of the 4 limbs (both at rest and in action), following the German-Swedish protocol. Spontaneous pathological postures were assessed by inspection (addressing equinus foot, hip endo-rotation/flexion, elbow flexion, abnormal posture of the shoulder, and impaired head and trunk control). Range of motion (ROM) was assessed in all flexion-extension-rotation directions normally possible in shoulder/elbow/wrist and hip/knee/ankle and graded by the clinician as not restricted/slightly restricted/obviously restricted. Fixed scoliosis and kyphosis was defined as a persistent spine deformity. Radiographic evidence of complete hip dislocation was always verified. The presence and severity of the motor impairments determined the most affected side of the body. In cases of a left-right symmetric presentation we included – arbitrarily – the right side of the body in further analyses.

Activity limitations were assessed according to the Dutch LIVRE system, a standardized recording system used at the time in all rehabilitation centres in the Netherlands (20). LIVRE is based on the SAMPC model addressing 5 activity domains, i.e. S: Somatic aspects; A: Activities of daily living; M: maatschappelijk (= social functioning in the community); P: psychological functioning (cognition and behaviour) and C: communication (21). In this study we focused on the first 2 areas, further indicated as mobility activities and self-care

activities, respectively. Mobility activities (10 items) included walking, rising, manipulating and lifting, self-care activities (3 items) refer to eating, toileting and washing (see Table III). The items were scored on a 4-point Likert scale describing difficulty of performance of the activity specified, with (0) indicating "manages without problems", (1) "slight difficulty, but manages", (2) "manages only with obvious difficulty or with help" and (3) "does not manage even with help". Sum scores of a domain were calculated, ranging from 0 to 30 (mobility activities) and 0 to 9 (self-care activities). Factor analyses confirmed unidimensionality of each domain (maximum likelihood, oblique rotation) with good reliability (Cronbach's alpha of 0.98 and 0.94, respectively). For further analyses item scores of ≥ 2 were indicated as an activity limitation; to dichotomize the sum scores we used the median score as cut-off point.

Analyses

Cases of missing data were negligible, since data collection took place by means of face-to-face interviews and physical examination. Anthropometric data (body weight, body height and BMI) were compared using the data-set from the Dutch Growth Foundation (22) on the Dutch child population in 1997 by means of 1-sample *t*-tests.

Associations between basic demographic characteristics (sex and age), CP characteristics (limb distribution of paresis, GMFCS and MACS level), and prevalence of motor impairments and limitations in activities were explored by Spearman's rank correlation coefficient. If less than 10% of the children suffered from a specific impairment, no correlation between impairment and CP characteristics was calculated. Additionally, we tested differences between subgroups of patients regarding limb distribution using the Pearson χ^2 -tests (in case of motor impairments) and analysis of variance (ANOVA) Tukey Honestly Significant Different *post hoc* tests (in case of activity limitations).

Table I. Characteristics of the children with cerebral palsy (CP)

Variable	Cohort (n=119)
<i>Child characteristics</i>	
Sex, male, n (%)	75 (63.0)
Age, years, mean (SD)	11.1 (3.6)
Length, m, mean (SD)	1.44 (.19)*
Weight, kg, mean (SD)	39.0 (16.8)*
Body mass index, kg/m ² , mean (SD)	18.1 (4.1)
<i>CP characteristics, n (%)</i>	
Limb distribution	
Unilateral spastic CP	48 (40.3)
Bilateral spastic CP	71 (59.7)
Diplegia	42 (35.3)
Tetraplegia	29 (24.4)
GMFCS levels	
I	31 (26.1)
II	46 (38.7)
III	10 (8.4)
IV	10 (8.4)
V	22 (18.5)
MACS levels	
I	23 (19.3)
II	55 (46.2)
III	23 (19.3)
IV	4 (3.4)
V	14 (11.8)
Intellectual functioning	
Normal	43 (36.1)
Learning disability	31 (26.1)
Mental retardation	45 (37.8)

*Significantly lower compared with age-matched peers.

GMFCS: Gross Motor Functioning Classification System; MACS: Manual Ability Classification System; SD: standard deviation.

In order to determine the association between motor impairments and activities, univariate logistic regression models were computed. Subsequently the significant variables from the univariate analyses ($p < 0.05$, 2-tailed) were applied to multivariate logistic regression models. These variables were entered as a single block into the regression equation. Nagelkerke R-square values were used to reflect the proportion of declared variance. Analyses were performed with SPSS 14.0.

RESULTS

Child and CP characteristics

Table I gives an overview of the group characteristics of the 119 children with spastic CP. Nearly two-third (75/119) of the patients were boys. Basic anthropometry revealed that these children had both a lower height than the reference population of Dutch children (height related to age ($t = -7.76$, $df = 110$, $p < 0.001$)) and a lower weight (weight related to age ($t = -4.62$, $df = 110$, $p < 0.001$)). This deviation from the reference population was larger in higher GMFCS levels. The BMI did not differ significantly from the reference population ($t = -1.3$, $df = 110$, $p = 0.194$).

Sixty percent of the children had bilateral spastic CP (Table I). Almost two-thirds of the children were independent walkers (GMFCS-levels I–II: 64.8%). The MACS distribution showed

that the same proportion of the children handled objects without help (MACS levels I–II: 65%) In addition, approximately two-thirds of the children had normal intellectual functioning or learning disabilities.

Limb distribution by GMFCS level correlated strongly (Spearman's $r = 0.78$, $p < 0.001$). The children with GMFCS level I have mostly unilateral spastic CP (90%), whereas in the higher levels bilateral spastic CP was almost exclusively present (7 out of 10 and 22 out of 22 for levels IV and V respectively). The correlation between GMFCS level and MACS level was Spearman's $r = 0.68$ ($p < 0.001$). The distributions of GMFCS and MACS levels are comparable, especially for the low levels (Tables I and II).

Motor impairments

Tables IIa and IIb summarize the occurrence of motor impairments (elevated muscle tone, spontaneous pathological postures, impaired trunk or head stability, ROM deficits of the extremities and spine deformities) in relation to limb distribution, GMFCS level and MACS level.

Overall, 91 children (76.5%) had no ROM deficits of the upper extremities. Thirteen children displayed 1, and 15 children 2 or 3 ROM deficits in the most affected arm. Similarly, 58 children had no lower extremity ROM deficits, 21 children had

Table IIa. Motor impairments in numbers (in total cohort of 119 children with spastic cerebral palsy (CP))

CP characteristic	Elevated muscle tone at rest*		Elevated muscle tone in action*		Spontaneous pathological postures				Head and trunk control impaired	
	Arm	Leg	Arm	Leg	Shoulder retraction	Elbow	Hip	Equinus	Head	Trunk
<i>Limb distribution</i>										
Unilateral (n=48)	16	16	29	31	3	16	7	13	–	–
Bilateral (n=71)	29	60	52	67	2	21	35	35	16	41
Diplegia (n=42)	10	30	24	39	2	7	17	15	2	18
Tetraplegia (n=29)	19	27	28	28	0	14	18	20	14	23
Spearman's r†	–	0.52	–	0.38	^	–	0.36	0.22	0.32	0.60
p-value		<0.001		<0.001			<0.001	0.015	<0.001	<0.001
<i>GMFCS</i>										
I (n=31)	9	9	20	21	1	8	2	5	–	–
II (n=46)	15	29	25	36	3	12	17	20	–	4
III (n=10)	–	7	5	9	–	2	6	4	–	6
IV (n=10)	5	10	9	10	1	3	5	5	2	10
V (n=22)	16	21	22	22	–	12	12	14	14	21
Spearman's r	0.24	0.50	0.25	0.32	^	0.17	0.37	0.31	0.56	0.78
p-value	0.008	<0.001	0.006	<0.001		0.066	<0.001	0.001	<0.001	<0.001
<i>MACS</i>										
I (n=23)	2	10	7	15	1	1	3	5	–	5
II (n=55)	18	29	34	43	2	16	16	18	1	2
III (n=23)	12	20	22	22	2	9	12	12	3	17
IV (n=4)	4	4	4	4	–	3	3	2	1	3
V (n=14)	9	13	14	15	–	8	8	11	11	14
Spearman's r	0.40	0.39	0.52	0.32	^	0.35	0.34	0.33	0.54	0.61
p-value	<0.001	<0.001	<0.001	<0.001		<0.001	<0.001	<0.001	<0.001	<0.001
Total n of children with this impairment	45	76	81	98	5	37	42	48	16	41

*Most affected side.

†Correlation between impairments by limb distribution (1=unilateral spastic CP, 2=bilateral spastic CP).

–: not significant; ^: correlation not computed (low number of prevalent cases); GMFCS: Gross Motor Functioning Classification System; MACS: Manual Ability Classification System.

Table IIb. Motor impairments in numbers (in total cohort of 119 children with spastic cerebral palsy (CP)) (contd)

CP characteristic	ROM deficits (one or more)			Spine deformities	
	Arm*	Leg*	Dislocated hip	Fixed scoliosis	Fixed kyphosis
<i>Limb distribution</i>					
Unilateral (n=48)	6	12	–	–	–
Bilateral (n=71)	22	49	4	9	7
Diplegia (n=42)	6	29	1	3	2
Tetraplegia (n=29)	16	20	3	6	5
Spearman's r^\dagger	0.21	0.43	^	^	^
<i>p</i> -value	0.020	<0.001			
<i>GMFCS</i>					
I (n=31)	2	5	–	–	–
II (n=46)	7	25	–	–	–
III (n=10)	–	6	–	2	1
IV (n=10)	6	9	2	1	1
V (n=22)	13	16	2	6	5
Spearman's r	0.42	0.43	^	^	^
<i>p</i> -value	<0.001	<0.001			
<i>MACS</i>					
I (n=23)	–	9	–	1	1
II (n=55)	8	21	–	–	–
III (n=23)	7	16	2	2	1
IV (n=4)	4	4	–	2	1
V (n=14)	9	11	2	4	4
Spearman's r	0.48	0.32	^	^	^
<i>p</i> -value	<0.001	<0.001			
Total <i>n</i> of children with this impairment	28	61	4	9	7

*Most affected side.

†Correlation between impairments by limb distribution (1=unilateral spastic CP, 2=bilateral spastic CP).

^ Correlation not computed (low number of prevalent cases).

GMFCS: Gross Motor Functioning Classification System; MACS: Manual Ability Classification System; CP: cerebral palsy; ROM: range of motion; –: not significant.

1, and 40 children 2 or 3 ROM deficits in (the most affected) leg. Limb distribution (unilateral vs bilateral spastic CP), gross motor functioning and manual ability showed moderate to good correlation with elevated muscle tone in the legs at rest, impaired head and trunk control and ROM deficits in the leg ($p < 0.001$). Limb distribution (also unilateral spastic CP vs BSCP), did not correlate with impairments in the upper extremity. The prevalence of other motor impairments correlated only poorly with limb distribution and gross motor functioning. More severely affected gross motor functioning correlated, as could be expected, with elevated muscle tone, spontaneous pathological postures and impaired trunk and head control.

Motor impairments were not related to sex and age group. Sub-group comparison analyses of children with BSCP revealed that children with tetraplegia significantly more often suffered from impairments in the upper extremities, equinus position, problems with head and trunk control compared with children with diplegia ($p < 0.05$). The presence of both elevated muscle tone in the legs and spontaneous pathological posture of the hips did not differ between children with tetraplegia and diplegia.

Activity limitations

Table III shows the degree of functional activity limitation by limb distribution and GMFCS and MACS levels. The specific activities in which the highest proportion of the children encoun-

ters limitations were walking outdoors and climbing stairs (40–41%). Regarding self-care activities, 35% of the children had limitations in toileting and washing or bathing. Bilateral limb involvement correlated to limitations in mobility (Spearman's $r = 0.70$, $p < 0.001$) and self-care activities (Spearman's $r = 0.50$, $p < 0.001$). Correlations with levels of gross motor functioning and manual ability ranged from $r = 0.70$ to $r = 0.88$, see Table III. ANOVA *post hoc* analyses showed that children with tetraplegia encountered more activity limitations than children with diplegia, who in fact experienced more limitations than children with hemiplegia ($p < 0.001$).

Associations between motor impairments and activity limitations

As presented in Table IV, limitations in mobility activities were associated with deficits in the lower limbs in univariate modelling, while self-care activities were constrained by impairments in both the upper and lower extremities ($p < 0.001$ to $p < 0.05$). Multivariate models included only the determinants that were significant in the univariate analysis, and demonstrated that elevated muscle tone, as such, was no longer a determinant of activity limitation, once the other motor impairments were taken into account. We found, however, that children with one or more ROM deficits or a pathological posture in the legs were 2–3 times more frequently limited in both mobility and self-care activities ($p < 0.001$ to $p < 0.01$).

Table III. Distribution of activity limitations, by gross motor functioning classification system (GMFCS), manual ability classification system (MACS) and limb distribution (n = 119)

Functional activity domains	GMFCS					MACS					Limb distribution		
	I n=31	II n=46	III n=10	IV n=10	V n=22	I n=23	II n=55	III n=23	IV n=4	V n=14	Unilateral n=48	Bilateral Diplegia n=42	Tetraplegia n=29
<i>Mobility*</i>													
From lie to sit	–	1	3	7	22	1	2	12	4	14	–	10	23
From sit to stand	–	3	8	10	22	6	4	15	4	14	–	16	21
Walking indoors	–	4	9	10	22	5	5	17	4	14	–	21	22
Walking outdoors	–	7	9	10	22	5	6	19	4	14	1	19	20
Walking stairs	–	7	10	10	22	6	6	19	4	14	–	21	22
Positioning	–	–	2	4	19	1	–	7	3	14	–	6	16
Manipulating	–	4	2	6	19	1	–	13	3	14	1	9	18
Endurance	1	2	3	2	14	2	2	4	2	12	1	5	9
Bending	–	2	4	7	20	1	1	14	3	14	–	6	13
Lifting	–	5	3	7	21	1	4	13	4	14	1	7	13
Sum score (SD)	1.1 (0–30)†	5.6 (4.6)	14.9 (4.0)	21.7 (4.3)	27.5 (3.5)	4.5 (6.1)	4.2 (4.8)	18.1 (6.7)	25.8 (5.0)	29.2 (1.5)	2.1 (3.0)	11.5 (7.8)	23.5 (8.6)
Spearman's <i>r</i>					0.88					0.72			0.70‡
<i>p</i> -value					<0.001					<0.001			<0.001
<i>Self-care*</i>													
Eating/drinking	–	3	1	4	19	1	2	7	3	14	1	8	18
Toileting	–	7	3	9	22	3	4	16	4	14	4	13	24
Washing/bathing	–	10	3	8	22	2	7	16	4	14	4	16	23
Sum score (SD)	0.4 (0–9)†	1.6 (2.4)	2.4 (2.4)	5.6 (2.1)	8.2 (1.1)	1.1 (1.8)	0.9 (1.7)	5.1 (2.5)	8.0 (1.4)	8.6 (0.6)	0.9 (1.7)	2.7 (3.0)	6.6 (2.9)
Spearman's <i>r</i>					0.74					0.70			0.50‡
<i>p</i> -value					<0.001					<0.001			<0.001

*Numbers refer to cases with a limitation in activity, i.e. obvious difficulty (requires assistance or major adaptations or completely incapable).

†Mean sum score (SD).

‡Correlation between domain score by limb distribution (1 = unilateral spastic CP, 2 = bilateral spastic CP).

SD: standard deviation; CP: cerebral palsy.

Table IV. Determinants of limitations in activities, dichotomized sum scores (cut-off point median score). Univariate and multivariate logistic regression analyses (n = 119)

Impairments†	Univariate analyses				Multivariate analyses			
	Mobility activities		Self-care activities		Mobility activities		Self-care activities	
	OR (95% CI)	R ²	OR (95% CI)	R ²	OR (95% CI)	R ²	OR (95% CI)	R ²
<i>Determinants</i>						0.42***		0.34***
Tonus‡								
Arm at rest	n.s.		3.1** (1.4–6.7)	0.09	–		n.s.	
Arm in action	n.s.		3.0** (1.3–6.8)	0.08	–		n.s.	
Leg at rest	8.9*** (3.6–22.1)	0.27	3.5** (1.6–7.9)	0.11	n.s.		n.s.	
Leg in action	8.0** (2.2–28.9)	0.15	2.9* (1.1–8.2)	0.05	n.s.		n.s.	
Spontaneous pathological postures								
Armδ	n.s.		2.5* (1.2–5.2)	0.07	–		n.s.	
Leg¶	3.2*** (1.8–5.7)	0.20	3.9*** (2.1–7.0)	0.25	n.s.		2.8*** (1.3–5.7)	
ROM deficits								
Arm§	2.2** (1.3–3.8)	0.11	2.4** (1.3–4.3)	0.12	n.s.		n.s.	
Leg°	3.1*** (2.0–4.8)	0.35	1.9*** (1.4–2.8)	0.16	2.0** (1.2–3.4)		n.s.	
Spine deformities♦	n.s.		n.s.		–		–	

p* < 0.05, *p* < 0.01, ****p* < 0.001.

†In limb of most affected side.

‡0 = not elevated; 1 = elevated.

δPresence of spontaneous pathological posture in elbow and/or shoulder (range 0–2).

¶Presence of spontaneous pathological posture in ankle and/or hip (range 0–2).

§ROM deficit of wrist and/or elbow and/or shoulder (range 0–3).

°ROM deficit of ankle and/or knee and/or hip (range 0–3).

♦Presence of fixed scoliosis and/or kyphosis (range 0–1).

OR: odds ratio; CI: confidence interval; R²: explained variance by Nagelkerke *R*-square test; –: not tested; n.s.: not significant.

DISCUSSION

The nature and prevalence of motor impairments and activity limitations in spastic CP has been studied in previous publications, originating from population-based studies or pooled populations (2, 4–6). The results of these studies share elements such as distributions of clinical presentations and GMFCS and MACS levels, but in their conclusions and considerations accents differ. For example Östensjö et al. (4) reported in 2004 that “spasticity and ROM deficits were both stated to be of importance for predicting functional performance along with selectivity of movement”; however, “motor impairments were only one component among many factors that could predict gross motor function and everyday activities”. This finding is confirmed in our study.

More recently in the Netherlands, longitudinal studies on the nature and course of motor impairments and activities in children and adolescents with CP have been and are being performed within the PERRIN programme (PEdiatric Rehabilitation Research In the Netherlands) (23, 24). PERRIN studies recruit their subjects via cooperating rehabilitation centres, thus focusing on a group within the CP population. CP (sub) groups that are followed longitudinally give good intra-subject and intra-group insight. The cross-sectional population-based study presented in this paper covers a wide field of aspects regarding CP children. Thus, in the Netherlands, cross-sectional “population-based” data join longitudinal “focused” data.

Children's characteristics

A general description of a representative group of 119 Dutch children with spastic CP is provided, as well as prevalence data on specific impairments and activities in mobility and self-care. The distribution of boys/girls and of major clinical characteristics is in line with other representative pooled data (1). Children with spastic CP had both a lower age-adjusted body height and body weight compared with typically developing peers (22); this was especially the case in children with higher GMFCS levels. This latter finding is in line with reports from multi-centre studies on growth in American children with moderate to severe CP (GMFCS III–V) (25). Children with spastic CP did not differ from the general population with respect to their BMI. Although CP is more prevalent in boys (1), we found that the consequences of CP in terms of the nature of specific motor impairments and performance of activities were not related to sex or to age-group.

ROM deficits were encountered in 60% of the children. This means that no less than 40% of the children with spastic CP (especially the children with low GMFCS levels) had no ROM deficits whatsoever. Either the natural course of spastic CP in these cases had not resulted in what professionals indicate as contractures, or preventive treatment had been completely effective. This is interesting, as the need to prevent or treat the unavailability of “contractures” in CP is stressed frequently. We find that the “need to treat” is probably less present in less severe cases of CP. Children with spastic CP in GMFCS and MACS levels I and II who use their extremities actively perform ROM exercises in a sufficient way.

Severe orthopaedic problems, such as complete hip dislocation or fixed spine deformities, were seen only in GMFCS IV–V levels. However, the prevalence of these severe orthopaedic problems was low; only 4 children had hip dislocation (and this only unilaterally). These low numbers are puzzling as the co-existence of severe CP and hip dislocation is reported frequently. In a recent English study, within a group of children with bilateral spastic CP and GMFCS level V (41 individuals), 50% had a hip dislocation by the age of 15 years (26). In an older population-based Swedish study, 75% of children with tetraplegic CP had hip dislocation and scoliosis (27). In recent years Boldingh et al. (28) examined 160 Dutch patients with severe tetraplegic CP, aged 16–84 years, and found “moderate” hip deformity in 41% and “severe” hip deformity in 29%. These last 2 studies focused on subjects with 4-limb involvement and a high GMFCS level (which is of course a selected group) and included (much) older patients who may not have had preventive treatment during their growth. An explanation might be that in the Netherlands the long-standing practice of radiographic monitoring of hip migration including timely conservative or operative measures results in low rates of complete hip dislocation. Another possibility is that the average age of our cohort is lower than the age at which dislocations become manifest. Due to the low number of hip dislocations in our cohort we cannot analyse the relation with age subgroups. The low prevalence of hip dislocation in the Dutch population could be the subject of further research.

As could be expected, more severely affected gross motor functioning (higher GMFCS levels) and total body involvement, such as in tetraplegia, correlated strongly with the degree of impairment present and the presence of limitations in activities. This finding is not surprising and is frequently reported (2, 4, 6, 24). GMFCS and MACS are known to correlate, as shown by Eliasson et al. (19) and in this study. However, the difference of distribution in the higher levels between GMFCS and MACS show that they do classify different types of activities.

Over 60% of the children could walk without assistive walking devices (GMFCS levels I–II). Thus, a typical child with spastic CP will be an independent walker rather than a wheelchair-user. This finding is consistent with European population-based CP-studies pooling data from more than 6000 children (29).

Some aspects of the study should be addressed

We assessed the motor impairments and activity limitations, as they were encountered in the group of children with spastic CP who were being treated according to professional standards in the Netherlands. Previous and current treatments and interventions (orthopaedic surgery, orthotics, medication) were known to us from the parents' interview. No causal relationships have been statistically explored in this study between the impairments found and specific previous interventions, such as anti-spastic medication, advanced spasticity treatment (these 2 were hardly present in this cohort) or orthotics and orthopaedic operations (which both were frequently encountered). Impairments can be both present or absent, either in the natural course of CP or when interventions are (repeatedly)

undertaken. Exploration causality between interventions and impairments would need a longitudinal study design, such as a Swedish longitudinal study with the focus on prevention of hip dislocation, showing that that implementation of a protocol with radiographic hip development follow-up and timely interventions resulted in less dislocations compared with a control group that lacked this approach (7).

The distinction between diplegia and tetraplegia is supposed to be mainly of clinical importance (clinicians “picture” a child from this type of description). It has been suggested to use only the term “bilateral spastic CP” or BSCP (for CP epidemiology), which has the benefit of avoiding debates where diplegia stops and tetraplegia starts, but also because the GMFCS level by itself describes functional performance and a limb-oriented classification could move to the background (8). We recognized the important main groups unilateral and bilateral spastic CP, but kept track of the leg-dominated and 4-limb dominated subcategories of BSCP – diplegia and tetraplegia (owing to the fact that we used the German-Swedish protocol). We found that diplegia and tetraplegia differed not only from an impairment point of view (as reported by Östensjö et al (4)), but also from an activity point of view, as the children with spastic diplegia had fewer limitations compared with the children with tetraplegia, in self-care activities but also in the broad domain of mobility activities, including positioning and manipulating. We conclude that besides the obvious relevance of the terms diplegia and tetraplegia to physicians treating individual children, these terms do refer to differences in activities, which, for a child with the upper extremities less affected, also seems logical.

Clinical practice often assumes a direct relationship between impairments and activity limitations. Indeed, univariate models showed that increased tonus, pathological postures and ROM deficits were related to limitations of the studied activities. However, multivariate relations between impairments and activities revealed that mainly ROM deficits in the lower extremity were related to mobility activities and spontaneous pathological postures were related to self-care. Because there were only modest associations between the presence of motor impairments and limitations in activities, a treatment that specifically targets motor impairments (such as disorders of muscle tone) may not be sufficient to achieve an enhancement of activities. Other aspects, such as environmental aspects or non-motor impairments, have to be taken into account. Moreover, what the ability to perform a given activity means to a child and his or her parents might be influenced by individual perceptions and expectations. For example, a child might prefer using a wheelchair with ease to walking, if walking is only possible with great effort using walking aids.

Limitations of the study

First there is a possibility of under-reporting of “minor however present” motor impairments and activity limitations as we chose to do the analyses with motor impairments that were more than “slight, or minor” and with activity limitation that were at least valued as “obvious”. Including the “slight” or “minor” category for both could give rise to threshold-issues

between “no problems” and “very minor problems” and the practical relevance may be limited.

The LIVRE method was designed in the 1990s as a registration tool and has not been validated, in the way the Pediatric Evaluation of Disability Inventory (PEDI) has been, which became available also in Dutch (30). LIVRE was in use in all Dutch rehabilitation centres at the time of the study and gave a bird’s-eye view of the patients’ functioning in 5 major domains of functioning. To date in 2008 the 5 SAMPC domains used in LIVRE are still the basis of many systematic medical patient files in rehabilitation medicine in the Netherlands. The 4-step LIVRE grading of none, minor, obvious difficulty up to impossibility to perform the activity resembles the result-oriented scoring of PEDI. Moreover, the risk that we indicated a limitation erroneously is low, since we started at obvious difficulty or worse as scores to indicate an activity limitation.

GMFCS and MACS were not available at the time the physicians examined the child. The classification was done on a retrospective basis by the first author. Comprehensive information from the parents on the child’s performance in daily life, assistive devices, the personal examination and an extra qualitative structured description of both walking and of hand and arm function yielded a vivid picture of the child. Both GMFCS and MACS are known to be rather unequivocal, use descriptions of the levels that also non-professionals can deal with, which contributes to the good inter-observer reliability (18, 19). So “knowing, examining and observing the child personally” formed a good basis for *post-hoc* classification. Borderline classification issues will always be present (and will be as well in the observations done today) but the contours of the different levels are clear-cut.

In conclusion, the distribution of CP-related characteristics in this Dutch cohort is consistent with that found in other representative studies. The prevalence of motor impairments and activity limitations has been determined in relation to major CP characteristics. A markedly low rate of hip dislocation was found in comparison to other studies. Distinction between diplegia and tetraplegia is relevant from a clinical point of view but also from an activity point of view. Activity limitations are determined only partly by the mere presence of motor impairments, which confirms the findings of other studies (4, 8). Individual goal setting in rehabilitation should identify all factors relevant to the child, including environmental factors. An activity-oriented rehabilitation approach goes beyond the treatment of motor impairments that are present.

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REFERENCES

1. Surveillance of Cerebral Palsy in Europe (SCPE). Prevalence and characteristics of children with cerebral palsy in Europe. *Dev Med Child Neurol* 2002; 44: 633–640.
2. Nordmark E, Hagglund G, Lagergren J. Cerebral palsy in southern Sweden II. Gross motor function and disabilities. *Acta Paediatr* 2001; 90: 1277–1282.
3. Östensjö S, Carlberg EB, Vollestad NK. Everyday functioning in young children with cerebral palsy: functional skills, caregiver assistance, and modifications of the environment. *Dev Med Child Neurol* 2003; 45: 603–612.
4. Östensjö S, Carlberg EB, Vollestad NK. Motor impairments in young children with cerebral palsy: relationship to gross motor function and everyday activities. *Dev Med Child Neurol* 2004; 46: 580–589.
5. Morris C, Kurinczuk JJ, Fitzpatrick R, Rosenbaum PL. Do the abilities of children with cerebral palsy explain their activities and participation? *Dev Med Child Neurol* 2006; 48: 954–961.
6. Beckung E, Hagberg G. Neuroimpairments, activity limitations, and participation restrictions in children with cerebral palsy. *Dev Med Child Neurol* 2002; 44: 309–316.
7. Hagglund G, Andersson S, Duppe H, Lauge-Pedersen H, Nordmark E, Westbom L. Prevention of dislocation of the hip in children with cerebral palsy. The first ten years of a population-based prevention programme. *J Bone Joint Surg Br* 2005; 87: 95–101.
8. Gorter JW, Rosenbaum PL, Hanna SE, Palisano RJ, Bartlett DJ, Russell DJ, et al. Limb distribution, motor impairment, and functional classification of cerebral palsy. *Dev Med Child Neurol* 2004; 46: 461–467.
9. Wichers MJ, Odding E, Stam HJ, van Nieuwenhuizen O. Clinical presentation, associated disorders and aetiological moments in cerebral palsy: a Dutch population-based study. *Disabil Rehabil* 2005; 27: 583–589.
10. World Health Organization (WHO). International Classification of Functioning, Disability and Health (ICF). Geneva: WHO; 2001.
11. Wichers MJ, van der Schouw YT, Moons KG, Stam HJ, van Nieuwenhuizen O. Prevalence of cerebral palsy in The Netherlands (1977–1988). *Eur J Epidemiol* 2001; 17: 527–532.
12. Mutch L, Alberman E, Hagberg B, Kodama K, Perat MV. Cerebral palsy epidemiology: where are we now and where are we going? *Dev Med Child Neurol* 1992; 34: 547–551.
13. Hagberg B, Hagberg G, Olow I. The changing panorama of cerebral palsy in Sweden. VI. Prevalence and origin during the birth year period 1983–1986. *Acta Paediatr* 1993; 82: 387–393.
14. Krageloh-Mann I, Hagberg G, Meisner C, Haas G, Eeg-Olofsson KE, Selbmann HK, et al. Bilateral spastic cerebral palsy – a collaborative study between southwest Germany and western Sweden. III: Aetiology. *Dev Med Child Neurol* 1995; 37: 191–203.
15. Krageloh-Mann I, Hagberg G, Meisner C, Schelp B, Haas G, Eeg-Olofsson KE, et al. Bilateral spastic cerebral palsy – a comparative study between south-west Germany and western Sweden. I: clinical patterns and disabilities. *Dev Med Child Neurol* 1993; 35: 1037–1047.
16. Krageloh-Mann I, Hagberg G, Meisner C, Schelp B, Haas G, Eeg-Olofsson KE, et al. Bilateral spastic cerebral palsy – a comparative study between southwest Germany and western Sweden. II: epidemiology. *Dev Med Child Neurol* 1994; 36: 473–483.
17. Hagberg B, Hagberg G, Olow I. The changing panorama of cerebral palsy in Sweden 1954–1970. I. Analysis of the general changes. *Acta Paediatr Scand* 1975; 64: 187–192.
18. Palisano RJ, Hanna SE, Rosenbaum PL, Russell DJ, Walter SD, Wood EP, et al. Validation of a model of gross motor function for children with cerebral palsy. *Phys Ther* 2000; 80: 974–985.
19. Eliasson AC, Krumlinde-Sundholm L, Rösblad B, Beckung E, Arner M, Ohrvall AM, et al. The Manual Ability Classification System (MACS) for children with cerebral palsy: scale development and evidence of validity and reliability. *Dev Med Child Neurol* 2006; 48: 549–554.
20. Goor GHW, editor. Landelijk informatiesysteem voor revalidatie (LIVRE). Nedrelandse vereniging van artsen voor revalidatie en fysische geneeskunde [Dutch society of physicians for rehabilitation medicine]. Instruction book. Utrecht: VRIN; 1995 (in Dutch).
21. Bangma B, Kap A. Inleiding revalidatiegeneeskunde [Introduction in rehabilitation medicine]. Assen: Van Gorcum; 1988 (in Dutch).
22. Groeistichting N. Growth Analyser Version 3.5. Rotterdam: In Edition; 2001–2006.
23. Donkervoort M, Roebroek M, Wiegerink D, van der Heijden-Maessen H, Stam H; The Transition Research Group South West Netherlands. Determinants of functioning of adolescents and young adults with cerebral palsy. *Disabil Rehabil* 2007; 29: 453–463.
24. Voorman JM, Dallmeijer AJ, Schuengel C, Knol DL, Lankhorst GJ, Becher JG. Activities and participation of 9- to 13-year-old children with cerebral palsy. *Clin Rehabil* 2006; 20: 937–948.
25. Stevenson RD, Conaway M, Chumlea WC, Rosenbaum P, Fung EB, Henderson RC, et al. Growth and health in children with moderate-to-severe cerebral palsy. *Pediatrics* 2006; 118: 1010–1018.
26. Morton RE, Scott B, McClelland V, Henry A. Dislocation of the hips in children with bilateral spastic cerebral palsy, 1985–2000. *Dev Med Child Neurol* 2006; 48: 555–558.
27. Edebol-Tysk K. Epidemiology of spastic tetraplegic cerebral palsy in Sweden. I. Impairments and disabilities. *Neuropediatrics* 1989; 20: 41–45.
28. Boldingh EJ, Jacobs-van der Bruggen MA, Bos CF, Lankhorst GJ, Bouter LM. Radiographic hip disorders and associated complications in severe cerebral palsy. *J Pediatr Orthop B* 2007; 16: 31–34.
29. Beckung E, Hagberg G, Uldall P, Cans C. Probability of walking in children with cerebral palsy in Europe. *Pediatrics* 2008; 121: e187–e192.
30. Wassenberg-Severijnen JE, Custers JW, Hox JJ, Vermeer A, Helden PJ. Reliability of the Dutch Pediatric Evaluation of Disability Inventory (PEDI). *Clin Rehabil* 2003; 17: 457–462.