

## MUSCULOSKELETAL PAIN IN ADULTS WITH CEREBRAL PALSY COMPARED WITH THE GENERAL POPULATION

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**Objective:** To examine prevalence and localization of musculo-skeletal pain in adults with cerebral palsy compared with the general population and to investigate variables potentially associated with pain.

**Design:** A postal survey.

**Subjects:** Persons with cerebral palsy and no intellectual disabilities, 18 years or more, living in Norway.

**Methods:** A multidimensional questionnaire, including items on musculo-skeletal pain, was sent to 766 adults with cerebral palsy.

**Results:** In total 406 persons responded, 49% females and 51% males age range 18–72 years (mean 34 years). All categories of cerebral palsy were represented. Nearly one-third of the adults with cerebral palsy had chronic pain, vs 15% in the general population. Mean scores of domain of bodily pain on Short Form 36 were significantly lower from an earlier age in adults with cerebral palsy. Back pain was the most common in both groups. Pain in adults with cerebral palsy was significantly associated with gender, chronic fatigue, low life satisfaction and deteriorating physical function.

**Conclusion:** Musculo-skeletal pain is a pronounced problem in adults with cerebral palsy from an early age, and should be addressed specifically in the follow-up programs, and in further clinical studies on potential causal pathways.

**Key words:** cerebral palsy, adults, musculo-skeletal pain.

J Rehabil Med 2004; 36: 78–84

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Submitted November 29, 2002; Accepted August 29, 2003

### INTRODUCTION

Musculo-skeletal pain is widespread in the general population of western countries, with impact on work, functional ability, and quality of life (1, 2). Although cerebral palsy (CP) is defined as a non-progressive brain lesion with primarily motor impairments (3), recent studies have described a gradual onset of new problems in adults with CP, such as musculo-skeletal pain, fatigue and deterioration of functional skills (4–16). However, it

is unknown whether the prevalence of pain in adults with CP is higher than in the general population. Previous studies on pain in adults with CP have not been performed with control groups or compared with reference groups (7–16). Given the high prevalence of musculo-skeletal pain in the general population, norm-based interpretation is of special interest. Thus, the present study has aimed to examine the prevalence and localization of perceived musculo-skeletal pain in adults with CP and to compare the results with those from similar studies in the general population of Norway. We also wanted to investigate and discuss the variables potentially related to musculo-skeletal pain in adults with CP.

### METHODS

#### *Participants and data collection*

The present study was based on a survey on adults with CP in Norway (4–6) including items on prevalence and localization of musculo-skeletal pain. Inclusion criteria were persons, 18 years or more, with diagnosed CP, living in Norway. Exclusion criteria were intellectual disabilities, documented in the medical records of potential participants. There is no central CP register in Norway, so adults with CP were not easily identified. Given that the prevalence of cerebral palsy in Norway coincides with other western countries, 2 per 1000 live births, the number of persons with CP in Norway is estimated to be approximately 6000 (17). According to international epidemiological studies on CP, about 60% are 18 years or more (3). International studies have estimated the prevalence of intellectual disabilities in persons with CP to between 30–50%, depending on the definition and the cut off of IQ (3). According to our inclusion criteria an estimation of the actual population in Norway counted about 2000 persons. Potential respondents were traced from different eligible sources, such as the archives of relevant hospitals and institutions and from the Norwegian CP-association. The result was 950 potential respondents. A pilot study was performed in August 1999 on 20 adults with CP, and in November 1999 a questionnaire was sent to 850 adults with CP from all over Norway, as 100 of the potential respondents had died. One written reminder was sent to the non-respondents in January 2000. The questionnaire was not included in the second mailing. Norwegian population studies on musculo-skeletal pain in general (1) and bodily pain as a part of the Short Form 36 (2), were chosen for comparison. These normative studies obtained population norms for the Short Form 36 and the Nordic Questionnaire on musculo-skeletal pain in Norway, respectively (1, 2). The present study was approved of the Regional Committee for Medical Ethics and the Norwegian Social Science Data Services.

#### *The questionnaire*

The questionnaire was multidimensional and developed in collaboration with a group of 3 adults with CP (4–6). Most of the items were identical to Andersson & Mattsson's questionnaire, used on adults with CP in Stockholm (7). In addition we used the standardized instruments listed below. Minor revisions of the questionnaire were made after the pilot

study. The final questionnaire consisted of the following groups of items:

- Demographic items including gender, age, family, education and employment.
- Diagnostic items including type of CP, classified according to Hagberg et al. (18). The persons with congenital ataxia were classified in the group with dyskinesia, and the persons with ataxic diplegia were classified in the group with diplegia. Severity of impairment was indicated by means of Short Form 36 (SF-36) Norwegian version 1.2 (19), domain of physical function. The SF-36 yields an 8-scale health profile, as well as summary measures with a scale from 0 to 100, 100 being the best health score. Validity and reliability have been tested in more than 45 countries as part of the International Quality of Life Assessment (IQOLA) Project (2, 19). Further items on changes in locomotion skills/use of wheelchair during life were also included. Physical and emotional role function, and general, and mental health were investigated by means of the SF 36.
- Items on experiences and needs regarding physiotherapy and physical activity (6).
- Prevalence of musculo-skeletal pain during the last month was investigated regarding intensity and impact on work and activities in daily life by means of SF 36, domain of bodily pain (2). Items on pain localization, frequency and duration, were also included, in addition to items on factors increasing or reducing the pain. These items were presented as multiple choices, with possibility to fill in more than one response alternative. They were not identical with the items of Natvig et al. (1), however, we chose the same cut off regarding the definition of chronic pain; daily pain for 1 year or more.
- Fatigue was investigated by means of The Fatigue Questionnaire (FQ) (20), which measures physical and mental fatigue during the last month, in addition to 2 items on duration and extent of fatigue. FQ has been well validated internationally, also in Norway (20). The sum score was dichotomized according to the definition of chronic fatigue (5, 20); scores above a cut off for 6 months or more.
- Life Satisfaction was investigated according to the global item of the Life Satisfaction Scale (21). This instrument, which was developed to evaluate rehabilitation processes, measures both global and domain specific life satisfaction, and has been well validated in Sweden (21).

#### Statistical analysis

Initial power calculations, based on confidence interval; assuming a 50% prevalence of pain in adults with CP and a confidence width of 0.1, estimated a required sample size of 385 persons. Level of significance was set at  $p < 0.05$ . The data were analysed with descriptive statistical methods (mean, SD), *t*-tests, and one-way ANOVA (continuous data) with Tukey corrections for multiple tests, Pearson chi square tests and logistic regression techniques (categorical data). The procedures in SPSS, version 10.0 and GraphPad.com were used in performing the analyses. In the questionnaires having 4 or less answers missing on FQ and SF-36, means of these items were filled in.

## RESULTS

### Sample characteristics and prevalence of chronic pain

The questionnaire was sent to 850 adults with CP, and 766 of these were included in the study. Due to wrong diagnosis, undocumented intellectual disabilities or unknown addresses 84 respondents were excluded. In total 406 persons (53%) responded, 49% females and 51% males, aged from 18 to 72 years, with a mean age of 34 years (SD 11.4 years). The non-respondents coincided well with the respondents both according to age, type of CP, and geographical distribution (4–6). There were 38% with hemiplegia, 36% with diplegia, 17% with dyskinesia and 8% with quadriplegia. Nearly 40% scored more

Table I. Age, gender, educational status, marital status, employment, prevalence of chronic pain (daily pain  $\geq 1$  year), chronic fatigue (dichotomized scores  $\geq 4$ ,  $\geq 6$  months) (Fatigue Questionnaire), (20), and global life satisfaction (dichotomized scores 4–6 vs 1–3), (21), in adults with cerebral palsy (CP) ( $n = 406$ )

Sample characteristics in adults with CP	<i>n</i> (%)
Age groups (18–72 years)	
<29 years	158 (39)
30–39 years	144 (35)
40–49 years	57 (14)
50–59 years	32 (8)
>59 years	15 (4)
Gender	
Males	209 (51)
Females	197 (49)
Education	
Primary school (9 years)	159 (39)
Secondary school (12 years)	144 (36)
University (>12 years)	103 (25)
Marital status	
Single/divorced/widowed	308 (76)
Married/cohabitant	98 (24)
Employment	
Paying job/self-employed	134 (33)
100% disablement benefit	188 (46)
Other source of income	84 (21)
Chronic pain and fatigue	
Chronic pain ( $n = 399$ )	111 (28)
Chronic fatigue ( $n = 403$ )	69 (17)
Life satisfaction	
High life satisfaction ( $n = 402$ )	222 (55)
Low life satisfaction ( $n = 402$ )	180 (45)

than 75 on SF 36, domain of physical function, indicating good physical function, 25% scored 51–75, 17% scored 26–50, and 21% scored less than 26.

Sample characteristics, such as gender, age, marital status, education, employment, prevalence of chronic pain, fatigue, and global life satisfaction are presented in Table I. Nearly all the respondents (92%) reported having had systematically follow-up from the health care system until the age of 18, with main focus on physiotherapy and training of motor function. After that age there are no systematic follow-up programs for persons with CP in Norway, except for persons with additional intellectual disabilities.

In total 327 persons (82%) with CP reported musculo-skeletal pain from at least 1 part of the body, while 72 persons (18%) reported no pain at all. In comparison 85% in the reference group (1) reported pain from at least 1 part of the body, while 15% reported no pain. There were 111 respondents (28%) who reported daily pain for 1 year or more, vs 15% in the reference group (1). Mean age of pain debut was 21 years (SD 11 years) in adults with CP. The prevalence of chronic pain in adults with CP increased from 18% under 30 years of age to 40% from 60 years of age, with no significant gender difference. In the reference group the prevalence of chronic pain increased from 8% in the 20–22 years old female cohort to 31% in the 60–62 years old female cohort, and from 5% in the 20–22 years male cohort to 21% in the 60–62 years cohort (1).

Table II. Musculo-skeletal pain sites in different types of cerebral palsy

Body regions	Diplegia % n = 125	Quadriplegia % n = 24	Hemiplegia % n = 112	Dyskinesia % n = 61	Total % n = 322	p-value
Back	64 <sup>1</sup>	67	47 <sup>1</sup>	67	59	<0.05
Neck	38 <sup>1</sup>	38	41 <sup>2</sup>	62 <sup>1,2</sup>	44	<0.05
Foot/ankle	54 <sup>1</sup>	42	38	33 <sup>1</sup>	44	<0.05
Shoulder	38 <sup>1</sup>	33	39 <sup>3</sup>	61 <sup>1,3</sup>	43	<0.05
Knee	41	58 <sup>1</sup>	38	26 <sup>1</sup>	39	<0.05
Hip	35	42	34	39	36	ns
Arm	22	29	31	33	28	ns
Head	25	8 <sup>1</sup>	24	41 <sup>1</sup>	26	<0.05
Other	6	4	4	10	6	ns

Post hoc tests of group comparisons between the types of CP.

<sup>1</sup> Significant difference in prevalence of pain ( $p < 0.01$ ).

<sup>2</sup> Significant difference in prevalence of pain ( $p < 0.05$ ).

<sup>3</sup> Significant difference in prevalence of pain ( $p < 0.01$ ).

Variables increasing the pain in adults with CP were reported to be overexertion in 230 persons (73%), inactivity in 82 persons (26%), and cold weather in 44 persons (14%). Variables reducing the pain were rest, reported by 161 persons (51%), physiotherapy by 154 persons (49%), medication by 111 persons (35%), movement by 89 persons (28%) and warm weather by 45 persons (14%).

#### Pain sites

Musculo-skeletal pain sites in different types of CP are presented in Table II. Neck pain, shoulder pain and headache were most prevalent in persons with dyskinesia. Back pain was more equally distributed, with the lowest prevalence in persons with hemiplegia. Foot and ankle pain was most prevalent in persons with diplegia, while pain in the knees was most prevalent in persons with quadriplegia. Pain in the hips and arms was quite equally distributed in the different types of CP.

Tests were performed to study whether pain in different body parts was related to gender and age in adults with CP. The prevalence of pain in women was significantly higher than in men regarding headache (36% vs 17%,  $p < 0.001$ ), back pain (65% vs 53%,  $p < 0.03$ ), and pain in the hips (42% vs 30%,  $p < 0.05$ ). In the reference group women also reported more pain than men, from all parts of the body, except for back pain (1). The following pain sites were significantly related to age in the adults with CP; neck pain ( $p < 0.001$ ), shoulder pain ( $p < 0.01$ ) and back pain ( $p < 0.05$ ), with a peak between 30 and 40 years. In the reference group back pain was equally distributed (approximately 50%) in all age groups, while headache was reduced and pain in the lower limbs increased with age (1). The adults with CP reported a tendency to increased headache with age, and reduced pain in the lower limbs. However, these changes were not statistically significant.

The majority of the 327 respondents with pain reported pain from more than 1 part of the body. In total 18% reported pain from 1 part of the body only, and 43% reported pain from 2–3 body parts. There were 42% who reported pain from 3–5 parts of the body, and 13% reported pain from more than 5 body parts. The range of painful body parts was from 1 to 9. Mean number of pain

sites was 3.6 in women and 2.9 in men. In the reference group the mean number of pain sites was 4.1 in women and 3.3 in men (1).

In addition to pain, most of the 406 respondents with CP reported reduced range of motion in 1 or more joints. Only 57 persons (14%) reported no reduced range of motion, corresponding with the respondents reporting no pain. There were 78 persons (19%) who reported 1 stiff joint, 83 persons (20%) reported 2 joints with reduced range of motion, 76 persons (19%) reported 3 stiff joints, 53 persons (13%) reported 4 stiff joints, and 59 persons (15%) reported 5 or more joints with reduced range of motion. Number of painful body parts was significantly associated with number of joints with reduced range of motion ( $p < 0.001$ ).

#### Bodily pain (SF 36) related to sex and age and multivariable analysis of chronic pain

Pain intensity and the impact of pain on work and activities in daily life during the last 4 weeks are presented in Table III. Nearly half the sample reported moderate to severe pain intensity, and one-third reported moderate to extreme impact of pain in daily life. Mean scale scores of domain of bodily pain on SF-36, related to sex and age groups in adults with CP, and a reference group (2), are presented in Table IV. Males with CP

Table III. Pain intensity and impact of body pain (SF 36) (19, on work and activities in daily life during the last 4 weeks in adults with cerebral palsy

Intensity of body pain (n = 398)	n (%)
None	113 (28)
Very mild	38 (10)
Mild	68 (17)
Moderate	112 (28)
Severe	53 (13)
Very severe	14 (4)
Impact of bodily pain (n = 390)	
Not at all	167 (43)
A little bit	106 (27)
Moderately	67 (17)
Quite a bit	30 (8)
Extremely	20 (5)

Table IV. Domain of body pain from SF-36 (19), mean scale scores (SD), by sex and age in adults with cerebral palsy (CP) and the general population of Norway (2) (higher scores indicate less pain)

Age groups	<40 years		>40 years		All	
	M	F	M	F	M	F
Body pain in adults with CP	68.7 <sup>1,2</sup> (27.1) n = 152	58.5 <sup>1,2</sup> (22.7) n = 147	69.2 <sup>2</sup> (25.1) n = 55	55.6 <sup>1,2</sup> (25.5) n = 48	69.0 <sup>1,2</sup> (28.2) n = 207	57.8 <sup>1,2</sup> (27.6) n = 195
Body pain in a reference group	81.5 (22.7) n = 483	78.2 (23.7) n = 506	74.0 (25.8) n = 632	69.0 (27.3) n = 666	77.2 (25.0) n = 1115	73.0 (26.6) n = 1172

<sup>1</sup>  $p < 0.001$  significant differences in mean scores of bodily pain between adults with CP and a reference group (2).

<sup>2</sup>  $p < 0.01$  significant differences in mean scores of bodily pain between males and females with CP.

had significantly less pain than females, and the same was the case in the reference group (2). The differences in bodily pain scores between the adults with CP and the reference group were statistically significant in males, 40 years of age or less. In females the differences in bodily pain scores were statistically significant in both age groups. Bodily pain was significantly associated with age in the reference group (2), but not in the adults with CP.

To investigate clinically interesting subgroups of persons with musculo-skeletal pain, we dichotomized the respondents into 2 groups, with and without long lasting, daily disabling pain. Simple and multiple logistic regression analyses were conducted to examine variables potentially associated with chronic pain. The results are presented in Table V. Age, low scores on physical function, physical and emotional role function, and life satisfaction were significantly associated with chronic pain in the univariate analysis, in addition to chronic fatigue, deterioration of functional skills, and lack of regular physical activity. When controlling for confounding variables in the multiple analysis, low score on physical role function, low life satisfaction, and deterioration of functional skills were significantly associated with chronic pain.

## DISCUSSION

The results of the present study showed that musculo-skeletal pain is a pronounced problem in adults with CP, with both similarities and differences compared with the general population. The prevalence of chronic disabling pain was significantly higher in the adults with CP than in the general population (1), and many of the persons with CP reported that pain had lasted since childhood. However, the prevalence of persons with no musculo-skeletal pain was similar in the adults with CP and in the reference group (1), also coinciding with the findings in other studies on adults with CP (7, 12, 13). However, it is difficult to compare the results of studies on pain in adults with CP, because of different functional status in the samples, different items in the assessment instruments and different definitions of chronic pain. The study by Schwartz et al. (11) defined chronic pain as daily pain with duration of 3 months or more, and in their study 56% fulfilled these criteria. In the study by Andersson & Mattsson (7) only 8% reported daily pain, but duration was not reported. In the present study chronic pain was defined as daily pain for 1 year or more, according to the population study by Natvig et al. (1), which

Table V. Variables potentially related to chronic pain (daily pain  $\geq 1$  year) in adults with cerebral palsy presented as odds ratio, 95% CI, and p-value. Coding of the categorical variables were: No chronic pain = 0 and chronic pain = 1 (1). Males = 0 and females = 1. No deterioration = 0 and deterioration = 1. Physical activity 60 min weekly or more = 0 and less physical activity = 1. High life satisfaction = 0 and low life satisfaction = 1 (21). No chronic fatigue = 0 and chronic fatigue = 1 (20). Age and the SF 36 variables (19), were treated as continuous. In the multiple regression analysis only the variables significantly associated with chronic pain are given (n = 111).

Variables	Univariate logistic regression (unadjusted)			Multiple logistic regression (adjusted)		
	Odds ratio	95% CI	p-value	Odds ratio	95% CI	p-value
Sex	1.32	0.85–2.05	ns			
Age	1.34	1.11–1.63	<0.01			
Education	1.28	0.76–2.15	ns			
Physical function (SF-36)	0.58	0.47–0.72	<0.001			
Physical role function (SF-36)	0.61	0.48–0.77	<0.001	0.74	0.55–0.99	<0.05
Emotional role function (SF-36)	0.67	0.47–0.96	<0.05			
General health (SF-36)	1.17	0.95–1.44	ns			
Mental health (SF-36)	1.07	0.69–1.66	ns			
Life satisfaction	2.94	1.86–4.65	<0.001	2.65	1.31–5.38	<0.01
Fatigue (FQ)	4.65	2.69–8.05	<0.001			
Deterioration of skills	5.57	3.25–9.54	<0.001	3.28	1.59–6.77	<0.001
Physical activity	1.90	1.21–2.99	<0.01			

gave a prevalence of 28%, nearly 100% higher than in the reference group.

Assumptions about different causes of pain cannot be addressed on the background of a cross-sectional survey. However, statistical analysis of variables potentially associated with musculo-skeletal pain, and other clinical studies, may contribute to generate hypotheses to guide further clinical research on causal pathways. Regarding pain distribution on body-parts, back pain was most frequently reported, both in the present and other studies on adults with CP (7, 11), and in the general population (1, 2). Approximately 50% of both genders in the reference group reported back pain, with no clear age differences (1), while in the present study back pain differed significantly with sex and age. There was also a tendency, however, not statistically significant, to reduced pain in the lower limbs with age, when many adults with CP start to use walking aids or a wheelchair. This might suggest that overuse could be a cause of pain in adults with CP. However, type of CP may be of greater importance regarding the prevalence of pain in different parts of the body than gender and age. CP is associated with musculo-skeletal deformities, such as subluxation and dislocation of the hip, abnormalities of the foot, patella alta, scoliosis, pelvic obliquity and contractures (22). In the present sample 86% reported reduced range of motion in 1 joint or more, and 18% reported contractures or deformities in 4 joints or more, corresponding significantly with the persons reporting chronic pain.

The distribution of pain sites in the different types of CP (Table II) showed that back pain, neck pain, shoulder pain and headache were considerably more prevalent in persons with dyskinesia than in the other groups. However, back pain had the same prevalence in persons with quadriplegia. It is well documented that repetitive involuntary movements and fluctuating tone in the trunk can create compressive and shearing forces causing cervical instability, disc herniation, spondylosis, osteophytes and stenosis of the spinal canal (23). Persons with quadriplegia are wheelchair users, and scoliosis is a common problem in this group, due to asymmetrical spasticity and immobilization (24). These findings indicate that both strain, caused by uncontrolled movements and immobilization might cause pain.

Pain in the ankles and feet was most prevalent in persons with diplegia, but also highly prevalent in persons with quadriplegia and hemiplegia. Nearly all persons with hemiplegia and about half of the persons with diplegia are walkers. However, persons with diplegia often walk with difficulty, and years of toe-walking, cavus foot deformities, or severe valgus/varus deformities can lead to pain in the metatarsal heads or on the medial or lateral border of the foot, respectively (22). Pain in the knees was most prevalent in persons with quadriplegia, but also highly prevalent in persons with diplegia and hemiplegia. Spasticity and immobility is commonly causing contractures and deformities in the lower limbs in the group with quadriplegia. Overactive quadriceps muscles might cause an overstretched patellar tendon with patella alta and chondromalacia, causing

pain, reduced strength and loss of function both in persons walking with flexed knees and in wheelchair users (25). This also indicates that immobilization as well as overexertion may increase musculo-skeletal pain. Pain in the hips, often caused by dislocation (26), was more equally distributed in the different groups, affecting both walkers and non-walkers. This tendency may also support the suggestion that both overuse and no use might cause musculo-skeletal pain.

Osteoarthritis is cited as one cause of pain in individuals with CP. Atypical joint compression from the imbalance of muscle activation across the joints can lead to degeneration and deformities of joint surfaces. Cathels & Reddihough (27) found arthritis in 27% of 149 subjects with CP. Osteoporosis, contractures and deformities also contribute to the risk of non-traumatic fractures in individuals with CP. Brunner & Doderlein identified 54 stress fractures in 37 persons with CP in a retrospective study of 20 years (28). The mean number of pain sites in the present study was 3 sites, 4 in women and 3 in men, confirming the results of other studies on both adults with CP and the general population (1, 2, 7, 11). These findings indicate that musculo-skeletal pain is mostly widespread pain, both in the general population and in adults with CP (1, 2, 7, 11).

Mean scores of bodily pain (SF 36) decreased significantly with age in the reference group (2), but not in the adults with CP (Table IV). The necessary changes of lifestyle that are reported by many of the adults with CP from before 40 years of age because of pain, fatigue and deterioration of function may explain this (4–6, 29). In general the mean scores in the adults with CP were significantly lower than in the reference group, except for males with CP from 40 years of age or more. This indicates that bodily pain in the adults with CP starts considerably earlier compared with the reference group, and consequently seems to represent phenomenon other than normal ageing. The results also indicate that the sum of factors that cause pain is influencing the development of pain differently in the genders.

Overexertion and weather changes were commonly reported factors increasing the pain, both in the present study and in the study by Schwartz et al. (11). In addition the present study reported immobility as a factor increasing the pain, while Schwartz et al. (11) reported depression as a factor increasing pain. Our study showed significant association between chronic pain and low scores on emotional role function (Table V). Both studies also pointed out the association between pain and fatigue. Rest, physiotherapy and staying active were common pain relievers in both studies, indicating that finding the balance between activity and rest is crucial. If musculo-skeletal pain has too high an impact on activities of daily life (Table III), many persons with CP decide to change their lifestyle, such as reducing their working hours, starting to use a wheelchair or other compensatory technical devices, and applying for increased home service (29). These changes may stabilize the pain, but they are often experienced as a defeat, causing serious psychological distress (4–16). A wheelchair is paradoxically looked upon as the ultimate enemy. However, our experience is,

that after having started to use one, most users ask themselves why they did not start earlier. In a 5-year follow-up study on polio survivors, Schanke (30) described how polio survivors experience similar psychological trauma as adults. However, after some years they found a new orientation in life and felt better.

Logistic regression analysis was conducted in order to identify clinically interesting subgroups regarding musculoskeletal pain. This analysis underlined the association between chronic pain, deterioration of functional skills, lack of adapted physical activity, low life satisfaction and low physical role function, which represents the impact of function on work and activities in daily life. Other studies (11, 12) have found low impact of pain on daily life in adults with CP, explained by a floor effect resulting from the already low activity level. However, the present sample represented all types of CP: one-third having paid work, and nearly half of the respondents being only mildly impaired. Jahnsen et al. (6) documented that severe impairment and lack of physical activity increased the odds ratio regarding chronic pain with 82% and 92%, respectively.

These results indicate that chronic musculo-skeletal pain is a pronounced problem in adults with CP compared with the general population, with negative impact on work and daily life, on the preservation of functional skills and on quality of life. Therefore pain should be addressed specifically in the follow-up programs for persons with CP from an early age. There is also a need for further clinical studies investigating the causes of musculo-skeletal pain in persons with CP, as they are exposed to more risk factors than the general population.

#### *Selection and recall bias*

Since there is no central CP-register in Norway potential participants were not easily identified, specially the oldest ones. The medical records from the actual hospitals and institutions cover mostly persons with CP born after 1960. However, 164 persons (104 respondents) older than 40 years were identified (4–6). Addresses, age, gender and diagnosis were described in the medical records, and this made us able to compare respondents and non-respondents according to these variables. As far as we know there is no systematic skewness between the respondents and the non-respondents of the present study, which gives our study a good internal validity.

However, as intellectual disabilities were an exclusion criterion, there are few respondents (33 persons) with quadriplegia, which is strongly related to intellectual disabilities. Some of them might even have a diplegia diagnosis today, as some studies say that quadriplegia is always related to intellectual disability (3). The lack of participants with high age and severe quadriplegia makes the external validity of the study weaker, according to the possibility of generalization. However, our sample seems to be fairly representative of adults with CP without intellectual disabilities.

Pain is a subjective and personal phenomenon, and as such, pain is best investigated by reports from the persons who experience it. However, in surveys there is always a risk that the

questions are misunderstood or interpreted differently by different persons. Perceptual problems may have increased this risk in the present study. The reports of pain in different parts of the body could have been more valid and reliable if we had used a pain-drawing. For instance, we do not know whether the respondents have low-back pain or pain in the upper back, or whether pain in the arms refers to elbows or wrists. Another problem was that approximately half of the respondents needed help in filling in the questionnaire. This may have influenced their answers, both regarding honesty and the use the caregivers' time.

Duration, intensity and the impact of pain in daily life may be biased by the difficulty in remembering things that happened long time ago, or the start of experiences that have been lasting for many years and become a habit. Mean duration of pain in the adults with CP was 13 years, indicating that many of the respondents have had musculo-skeletal pains since childhood. Self-reported symptoms are also a question of culture. The results of surveys on health problems will always reflect differences in judgement of what symptoms ought to be reported (1, 2). This phenomenon may give both under- and over-estimation of prevalence. Minor periods of pain may have been forgotten or neglected, and the pain may have become a more or less conscious habit. The choice of valid and reliable assessment tools hopefully have reduced these problems, so that the present survey may contribute to point out directions for further clinical studies, where potential causal pathways and treatment of musculo-skeletal pain in adults with CP could be addressed.

#### ACKNOWLEDGEMENTS

The present study was financially supported by Sunnaas Rehabilitation Hospital and by the Norwegian CP-Association. We would like to thank the 406 adults with CP who participated in the study.

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