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What is This?

Locomotion skills in adults with cerebral palsy

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Objectives: To investigate self-reported locomotion skills in persons with cerebral palsy (CP) and to investigate variables potentially associated with deterioration of walking skills.
Design: Cross-sectional retrospective survey.
Setting and subjects: A multidimensional mailed questionnaire was sent to 766 persons with CP, 18 years or over, without intellectual disabilities, living in Norway.
Main outcome measures: The questionnaire consisted of demographic and diagnostic items, items on locomotion skills, and physical function (SF-36).
Results: In total 406 persons, 51% males and 49% females from 18 to 72 years (mean 34 years, SD 11 years) with all categories of CP responded. Median age for reported walking debut was 3 years, with a range from 1 to 14 years. In total 216 respondents (53%) walked without support, 104 persons (25%) walked with support, 39 persons (10%) had lost their walking skills, and 47 (12%) had never been able to walk. Mean level of physical function (SF-36) was 53 out of 100. There were 97 persons (27%) who reported improvement of walking skills, mainly before 25 years, 102 (28%) reported no change, and 160 (44%) reported deterioration, mainly before 35 years of age. Deterioration was significantly associated with older age, delayed walking debut and severe neurological impairment. Self-reported causes of deterioration were pain, fatigue and lack of adapted physical activity.
Conclusion: Deterioration of locomotion skills is a significant problem in persons with CP from an early age, documenting the need for life-long follow-up. The predictors above should be investigated in further clinical studies, searching for potential causal pathways.

Introduction

Traditional follow-up programmes for children with cerebral palsy (CP) have to a large extent been oriented towards the development of motor skills, especially the ability to walk.[1,2] Independent walking has a very strong practical and symbolic value for children with CP, as well as for their parents and health professionals, and is often a main goal in treatment programme. This approach typically does not consider how much time and energy it takes to reach this goal, or how long the acquired skills are maintained.[1,2] A wheelchair has been looked upon as an antagonist to independent walking instead of a tool to save energy and maintain walking for as long as possible.

Recent studies have described the gradual onset of new impairment problems in adults with CP,

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such as chronic fatigue, musculoskeletal pain and deterioration of functional skills, with consequences for activities and participation in work and social life.[1–15] The consensus definition of CP by Mutch *et al.*[16] also points out that the consequences of CP change over time, although the brain lesion is considered to be nonprogressive. Nearly all elderly people in the general population experience some deterioration of functional skills.[17] Studies on adults with CP, however, show that they experience such deterioration from adolescence and early adulthood.[1–15] The causes of early deterioration are not yet known, but the studies mentioned above hypothesize that ageing, immobilization and overuse of the nervous system, muscles and limbs affected by chronic disablement might form a vicious circle, causing the new secondary problems.

In other published parts of our survey,[2,4,5] musculoskeletal pain and fatigue in adults with CP compared with pain and fatigue in the general population are addressed and discussed in relation to physical activity. Coping potential regarding these unexpected new challenges is also addressed.[3] The present part of the survey examined self-reported locomotion skills in persons with CP, including walking debut, walking distance, use of walking aids, and experiences and time of onset of secondary limitations of locomotion skills. In addition we wanted to describe self-reported thoughts about causes of deterioration, and investigate and discuss variables potentially predicting these new functional problems, in order to guide further clinical studies on causal pathways.

Methods

Participants and data collection

The present study was based on a retrospective survey on adults with CP in Norway,[2–5] and the data collection is described in more detail in other parts of the survey.[2,3] There is no central CP register in Norway. However, we obtained access to the diagnostic archives of Sunnaas Rehabilitation Hospital and Rikshospitalet University Hospital, Section for Child Neurology in Oslo, and the Norwegian CP Association also contributed. A pilot study was performed in August 1999, and in November 1999 a multidimensional questionnaire

was sent to 850 adults over 18 with CP with no intellectual disabilities documented in the medical records from all over Norway. One written reminder was sent to the nonrespondents in January 2000. The survey was approved by the Regional Committee for Medical Ethics and the Norwegian Social Science Data Services.

The questionnaire

The questionnaire of the survey was multidimensional and developed in collaboration with a group of three adults with CP,[2–5] building on the questionnaire used by Andersson and Mattsson.[6] Minor revisions of the questionnaire were made after the pilot study, and the final questionnaire consisted of the following groups of items in this part of the survey:

1) Demographic items
2) Diagnostic items: type of CP,[18] level of physical function (Short Form 36)[17]
3) Items on locomotion skills[6]

Type of CP was classified according to Hagberg.[2–5,18] When the respondents did not know what type of CP they had, this was controlled in their medical records, if accessible. Data on walking skills were also checked for these respondents. Level of physical function was measured by means of the SF-36, Norwegian version 1.2 domain of physical function.[17] This domain investigates gross motor activities in daily life during the last four weeks, activities in daily life, and locomotion skills, such as walking from 100 m to more than 2 km, climbing stairs, and running or performing sports.[17] However, the functional limitations for nonwalkers are not described. Therefore items such as age of walking debut, walking distance, use of walking aids or wheelchair, improvement or deterioration of walking skills based on Andersson and Mattsson's questionnaire[6] were used.

Statistical analysis

The data were analysed with descriptive statistical methods, and univariate and multiple logistic regression analysis in SPSS, version 10.0. Comparison between data in the adults with CP and the normative data was performed with independent sample *T*-tests in www.GraphPad. Univariate and

multiple logistic regression were used to investigate variables potentially associated to deterioration of locomotion skills. The items on level of physical function from the SF-36, Norwegian version 1.2[17] are scaled from 1 to 100 with 100 as the best score of function. In the questionnaires having 4 or fewer items missing, means of these items were filled in.

Results

Sample characteristics

In total 766 adults with CP were included in the study, and 406 persons (53%) responded, 49% females and 51% males aged from 18 to 72 years, with a mean age of 34 years (SD 11 years). The whole country was represented. General sample characteristics are presented in previous articles by Jahnsen et al.[2–5] These also include a comparison of the respondents and the nonrespondents regarding all eligible parameters, such as geographical distribution, different types of CP, age and gender. The comparison showed that there were only minor differences between the two groups.[2–5]

Locomotion skills

The age of walking debut in children with different types of CP is presented in Table 1. There were 302 respondents (74%) who reported the age at which they learned to walk, and this was checked in the medical records for 79 respondents (20%), as these records were accessible to us. The reported age of walking debut was corrected according to the medical record for one person only, and for five of these respondents data on walking skills were not found. The majority learned to walk during preschool age. The 57 missing reports (14%) were not found in the medical records, or these records were not accessible to us. Median age of walking debut was 3 years for the whole group, 2 years in the group with hemiplegia, 4 years in the group with diplegia and 5 years in the group with dyskinesia.

Data on walking distance and use of wheelchair are presented in Table 2. More than half of the sample were able to walk without support for 1 km or more, never using a wheelchair. Only 22% always used a wheelchair, and the remaining respondents walked with support or alternated between walking and using a wheelchair.

Reported changes in the ability to walk in the different types of CP, and the time of onset of these changes, are presented in Table 3. Nearly one-third of the respondents reported improvement in their walking skills, mainly before the age of 25 years, and about the same number reported no change. The remaining respondents reported deterioration, mainly between 15 and 35 years of age.

Deterioration of walking skills in different age groups of adults with CP is presented in Table 4. The results show that the percentage of persons

Table 1 Age when learning to walk in different types of CP

Age (years)	Hemiplegia n	Diplegia n	Quadriplegia n	Dyskinesia n	Unknown n	Total n	%	Cumulative %
< 2	56	11	1	7		75	19	25
2–3	42	23	1	8	1	75	19	50
3–4	17	13	1	6	1	38	9	62
4–5	6	19	2	4	1	32	8	73
5–6	4	15	1	8		28	7	82
6–7	2	10	2	5		19	5	88
7–8	1	13	1	4	1	20	5	95
8–9	1	6	1	2		10	3	98
9–15		2		3		5	1	100
Total walked	129	112	10	47	4	302	74	
Never walked	2	9	21	15	0	47	12	
Missing	23	24	2	7	1	57	14	
Total	154	145	33	69	5	406	100	

Table 2 Self-reported walking skills and use of wheelchair in adults with different types of CP

Characteristics	Hemiplegia n	Diplegia n	Quadriplegia n	Dyskinesia n	Unknown n	Total n	%
Walking without support – total	128	56	0	27	5	216	53
< 50 m	0	0	0	0	0	0	–
50–199 m	0	2	0	0	0	2	–
200–499 m	1	3	0	2	0	6	–
500–1 km	4	0	0	2	0	6	–
> 1 km	123	51	0	23	5	202	–
No support inside – support outside	15	26	0	10	0	51	12
Walking with support – total	6	38	2	7	0	53	13
< 50 m	2	10	0	3	0	15	–
50–199 m	0	7	2	1	0	10	–
200–499 m	0	10	0	2	0	12	–
500–1 km	1	3	0	1	0	5	–
> 1 km	3	8	0	0	0	11	–
Not walking – total	5	25	31	25	0	86	22
Stopped walking	3	16	10	10	0	39	–
Never walked	2	9	21	15	0	47	–
Total	154	145	33	69	5	406	100
Use of wheelchair							
Never	137	65	0	24	5	231	57
Very seldom	6	13	0	3	0	22	5
Both walk and use wheelchair daily	6	41	2	17	0	67	16
Always	5	26	31	25	0	86	22
Total	154	145	33	69	5	406	100

reporting deterioration is higher in the older than in the younger age groups.

Physical function, including locomotion skills, related to gender and age in adults with CP was investigated according to the domain of physical function in SF-36. Males had a mean score of 55, with mean score of 62 below 30 years of age and of 23 after 50 years of age. Females had a mean score of 50, with mean score of 57 below 30 years of age and of 35 after 50 years of age.

The respondents reported that pain, fatigue and lack of adapted physical training created a vicious circle, causing the deterioration of locomotion. Deterioration was described as increasing balance problems in 93 persons (63%), reduced muscle strength in 62 persons (42%), endurance in 61 persons (42%), or combinations of symptoms, causing reduced walking speed and walking distance, and increased need for support or a wheelchair in the community. To identify variables potentially related to deterioration, the association with the age, age of walking debut, and type of CP was investigated. The results, based on univariate and multiple regression analysis, are presented in Table 5. Odds ratio regarding deterioration was significantly higher in the groups with severe type of CP and the groups with high age.

Discussion

Since there is no central CP register in Norway, older people with CP were difficult to identify. Intellectual disabilities were exclusion criteria and therefore there are few respondents with spastic quadriplegia, which is strongly related to intellectual disabilities. The limited number of older participants with severe quadriplegia affects the external validity of the study regarding the possibility of generalization. However, there were only minor differences between the respondents and the nonrespondents, which gives our study a good internal validity. The representativeness of our

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Clinical messages

- Deterioration of locomotion skills is a pronounced problem in adolescents and adults with cerebral palsy.
- Self-reported causes of deterioration are pain, fatigue and lack of adapted physical activity, while strong predictors are severe neurological impairment and older age.
- Adults with CP need systematic lifespan follow-up.

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sample is discussed in more detail in other parts of the survey.[2–5]

Subjective experiences, such as deterioration of function are best investigated by reports from the persons who experience them. However, there is always a risk that the questions in surveys are misunderstood or interpreted differently by different persons, and the answers will always reflect differences in judgement of what symptoms ought to be reported.[17] Perceptual problems and the need for help to fill in the questionnaire may have increased this risk and influenced the answers of the present study.[2–5] Experiences dependent on memory may also be biased. Events that happened a long time ago may be forgotten. Minor losses of function may have been neglected or become an unconscious habit. On the other hand, minor losses of function may have been enlarged by

Table 3 Self-reported changes of locomotion skills in adults with CP. The respondents who have stopped walking completely are a subgroup of those who have deteriorated

Characteristics	Hemiplegia n	Diplegia n	Quadriplegia n	Dyskinesia n	Unknown n	Total n	%
Walking skills (n = 406)							
Improved	53	30	1	11	2	97	27
Unchanged	55	33	1	11	2	102	28
Deteriorated	44	73	10	32	1	160	45
Total	152	136	12	54	5	359	100
Never walked	2	9	21	15	0	47	12
Total	154	145	33	69	5	406	100
Improvement – age (n = 97)							
< 15 years	32	12	0	6	2	52	54
15–24 years	15	12	1	4	0	32	33
25–34 years	2	3	0	1	0	6	6
35–45 years	0	3	0	0	0	3	3
> 45 years	1	0	0	0	0	1	1
Missing	3	0	0	0	0	3	3
Total	50	30	1	11	2	97	100
Deterioration – age (n = 160)							
< 15 years	5	4	3	3	0	15	9
15–24 years	12	22	5	6	0	45	29
25–34 years	17	31	0	9	0	57	35
35–45 years	5	9	1	9	0	24	15
> 45 years	5	7	1	5	1	19	12
Total	44	73	10	32	1	160	100
Stopped walking – age (n = 39)							
< 15 years	1	2	3	2	0	8	21
15–24 years	1	4	1	3	0	9	23
25–34 years	0	5	4	1	0	10	26
35–45 years	0	3	1	2	0	6	15
> 45 years	1	2	1	2	0	6	15
Total	3	16	10	10	0	39	100

Table 4 Deterioration of walking skills related to age groups in adults with CP

Age groups	No deterioration n (%)	Deterioration n (%)	Total n (%)
< 25 years	63 (80%)	16 (20%)	79 (100%)
25–34 years	78 (59%)	54 (41%)	132 (100%)
35–44 years	40 (45%)	48 (55%)	88 (100%)
≥ 45 years	18 (39%)	42 (70%)	60 (100%)

strong frustration. However, such recall bias may give both under- and overestimation of prevalence, and probably do not cause systematic skewness of the data in a large sample. In about 20% of the sample we were able to check on the diagnosis and data on walking skills in the medical records to validate our data, and the data on walking skills coincided well with the reported data. Since walking tended to be a main focus in the treatment of children with CP,[2] most persons who achieved independent walking would probably know when it happened. Most of our items had been used and found adequate for adults with CP in Stockholm,[6] and this has also hopefully reduced the bias problems.

The percentage of persons reporting improvement or deterioration of walking skills and the time of onset of the deterioration were similar in the present and other studies.[1,6,7] Willner and Dunning[13] and Murphy *et al*.[11] reported a higher prevalence of deterioration. However, their samples had higher prevalence of moderate and severe neurological impairment,[9,11] and higher mean age than the other studies.[9] Comparison of the results are difficult because of different descriptions and interpretations of the word walking. This reduces the validity of the data. Another weakness is that we only know the level of walking skill at the time of the assessment, and not when it was at its best. There is a need for a study describing the amount of deterioration. The use of an instrument like the Gross Motor Function Classification System (GMFCS)[19] with five levels of gross motor function could have described the amount of deterioration for instance, deterioration from level II to level III. However, this instrument has not yet been validated for adults, and has only been explored briefly in another part of our survey.[2] A follow-up study on validity and reliability of GMFCS in adults with CP is in progress at our hospital.

Nearly all elderly people in the general population experience some deterioration of locomotion skills.[17] The level of physical function in adults with CP was also lower in the older age groups. In the reference group, however, the level of physical function was relatively stable until after 60 years of age[17], while the marked differences in physical function in different age groups of adults with CP were seen from 30 years of age. Further clinical studies with longitudinal design are needed to

Table 5 Variables potentially related to deterioration of locomotion skills presented by odds ratio, 95% CI and p–value

Variables	Univariate logistic regression			Multiple logistic regression		
	Odds ratio	95% CI	*p-value*	Odds ratio	95% CI	*p-value*
Age						
25 years	1		< 0.001	1		
25–34 years	2.73	1.52–5.22	0.002	3.18	1.48–6.85	0.003
35–44 years	4.73	2.37–9.43	< 0.001	5.36	2.39–12.02	< 0.001
≥ 45 years	9.19	4.22–20.01	< 0.001	7.92	3.23–19.42	< 0.001
Age of walking debut	1.24	1.11–1.38	< 0.001	1.09	0.96–1.24	0.170
Type of CP						
Hemiplegia	1			1		0.011
Diplegia	2.84	1.75–4.65	< 0.001	1.96	1.06–3.62	0.031
Quadriplegia	12.27	2.58–58.28	0.002	8.38	1.49–47.08	0.016
Dyskinesia	3.57	1.87–6.81	< 0.001	2.76	1.25–6.08	0.012

The coding of the categorical variables (required to interpret the odds ratios) was: No deterioration= 0, deterioration= 1. The different types of CP are coded from 1 to 4. Age was categorized into four decades 1–4. Age of walking debut was treated as a continuous variable.

explain this, since a cross-sectional study cannot explain development of function.

As shown in the logistic regression analysis, deterioration of walking skills was most strongly associated with spastic quadriplegia, which is the most severe type of CP, and least with hemiplegia, which is the mildest type of CP. Older age was also strongly associated with deterioration. Ando and Ueda[7] reported similar results. This may indicate that ageing is a part of the deterioration process, as in the general population.[17] However, severity of impairment was an equally strong predictor. Deterioration of locomotion was significantly associated with age of walking debut in the univariate analysis. However, age of walking debut is also an expression of severity of impairment, contributing to confirm the importance of severity of impairment as a predictor of deterioration. This may support Bottos and coworkers' critique of the often one-sided emphasis on the training of independent walking in the follow-up programmes of children with CP for many years.[1] Training of walking skills was reported up to 14 years in our survey.[2]

Self-reported causes of deterioration were pain, fatigue and lack of adapted physical activity. Other parts of the survey showed association between musculoskeletal pain, fatigue and deterioration.[3,4] Immobilization seemed to be a larger problem than overuse, even though overuse syndromes probably are related to some subgroups of CP.[3] Chronic fatigue was most prevalent in respondents with moderate motor impairment, indicating that there may be an imbalance between 'work-load' and 'work-capacity' in the daily life of this group.[4] Our article on the relation between physical activity and the secondary complications in adults with CP[2] showed that the odds of early deterioration were reduced in the physically active. Improvement was reported to be due to increased adapted physical training, which supports this hypothesis. However, improvement was only related to young age. In 10 years these respondents might experience deterioration. Pimm[14] hypothesized a theory of overuse and 'physiological burnout', causing the new secondary problems. However, the present and other studies have shown significant association between deterioration and severe neurological impairment, which is related to inactivity.[15] Further clinical studies are needed to investigate potential causal pathways of the early deterioration in persons with CP.

Conclusions

The present study documents that deterioration of locomotion skills is a far more pronounced problem in adults with CP from an earlier age than the deterioration following ageing in the general population. Important predictive variables in relation to deterioration were severe neurological impairment and older age. Self-reported causes of deterioration were pain, fatigue and lack of adapted physical activity. The knowledge is still limited about whether the early deterioration is caused by overuse, inactivity or both, and there is a need for further clinical studies to search for potential causal pathways. Our findings may point out directions for further studies. The present study also underlines the need for lifespan follow-up programmes for persons with CP developed to meet the specific risk factors in the different subgroups of CP, overuse syndromes in some groups and inactivity syndromes in other groups. Therefore the follow-up programmes need to address the balance between physical activity and fitness on one side and saving of energy for personal pursuits on the other.

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