Received: 13 March 2019 | Revised: 23 May 2019 | Accepted: 18 June 2019

DOI: 10.1111/apa.14909

REGULAR ARTICLE

ACTA PÆDIATRICA NURTURING THE CHILD WILEY

Back pain is more frequent in girls and in children with scoliosis in the context of cerebral palsy

Gunnar Hägglund[1] | Tomasz Czuba[2] | Ann I. Alriksson-Schmidt[1]

[1]Department of Clinical Sciences Lund, Skåne University Hospital, Lund University, Orthopedics, Lund, Sweden

[2]Department of Clinical Sciences Lund, Lund University, Lund, Sweden

Correspondence
Gunnar Hägglund, Department of Clinical Sciences, Lund University, Orthopaedics, 221 85 Lund, Sweden.
Email: gunnar.hagglund@med.lu.se

Funding information
Nordforsk, Grant/Award Number: 82866; Forte and the Swedish Research Council, Grant/Award Number: 218-01468; Linnea and Josef Carlsson Foundation; Norrbacka Eugenia-stiftelsen; Jerringfonden and Stiftelsen för bistånd åt rörelsehindrade i Skåne

Abstract

Aim: To investigate the prevalence of general and back pain in children with cerebral palsy and the relationships between scoliosis and back pain.

Methods: Cross-sectional register study based on data from the Swedish Cerebral Palsy Follow-Up Programme. Descriptive analyses and logistic regression to regress age, sex, gross motor function, windswept, hip extension and source of report on the presence of pain.

Results: The study included 3783 children (58% boys) 1-18 (mean 10.0) years of age. General pain was reported in 1538 (44% girls, 38% boys) and back pain in 226 (7% girls, 5% boys) children. The proportion of back pain increased from <4% prior to age 12 years to >12% from 16 years of age. Back pain increased from 4% in children without scoliosis to 16% in children with severe scoliosis. Moderate/severe back pain increased from 2% in children without scoliosis to 10% in children with severe scoliosis. Increased odds of reporting back pain were found for age, girls, low gross motor function and children with scoliosis.

Conclusion: The proportion of children with general pain increased with age and was more frequent in girls. Age, female sex, low gross motor function and scoliosis were significant predictors of back pain.

KEYWORDS
cerebral palsy, children, pain, scoliosis

1 | INTRODUCTION

Cerebral palsy (CP) is caused by brain damage occurring in utero or prior to age 2 years and is one of the most common musculoskeletal disabilities in childhood.[1] CP results in a variety of manifestations; however, movement is always affected to some degree. CP might also result in activity limitations, and at times disturbances in communication, cognition, perception and sensation.[1] Given that gross motor function is central in CP, the gross motor function classification system (GMFCS) has been developed, with five ordinal and mutually exclusive categories (I-V), where GMFCS V indicates the most severely affected gross motor function.[2] GMFCS levels generally do not fluctuate much on an individual basis but tend to stay stable.[3,4]

Individuals with CP are at risk of developing neuromuscular scoliosis.[5] There are numerous problems associated with scoliosis including trunk decompensation, poor sitting and an increased risk of excessive skin pressure. Age and GMFCS level are predictors of the development of scoliosis in that the risk of scoliosis increases with

Abbreviations: CI, confidence interval; CP, cerebral palsy; CPUP, cerebral palsy follow-up programme; GMFCS, gross motor function classification system; OR, odds ratio; PT, physiotherapist.

GMFCS level and age.[5] Children at higher GMFCS levels (ie more severely affected gross motor function) have also been reported to develop scoliosis at an earlier age.[5] Additional risk factors for scoliosis include being female, having undergone hip surgery, moderate to severe epilepsy, poor muscle control, muscle weakness and spasticity.[6-9] Treatment modalities for scoliosis can be either surgical (segmental spinal fusion) or nonsurgical (spinal bracing).[10] Although surgery appears indicated for deformity correction, in a recent review article, insufficient evidence in terms of functional outcomes, caregiver outcomes and quality of life were reported.[10]

In addition to scoliosis, pain is a common secondary condition in CP, which, similarly with scoliosis, increases with age and is more frequently reported in those at higher GMFCS levels and in girls and women.[11-13] The complications of scoliosis can sometimes be painful. However, to the best of our knowledge the relationship between scoliosis and back pain has not been studied previously.

The aims of this study were to[1] investigate the prevalence of pain in general and back pain specifically in children and adolescents with CP in relation to scoliosis,[2] explore associations between back pain and scoliosis by age, sex, GMFCS level, self- versus proxy reporting and[3] assess the relationships between scoliosis severity and pain intensity level.

Key notes

- We assessed the prevalence of pain in general and back pain specifically in 3783 children and adolescents with cerebral palsy (CP) using Swedish register data.
- Pain was reported in 41% of all children aged 1-18 years, more frequently in girls and in the older children.
- Back pain was reported in 6% of all children and increased with degree of scoliosis up to 16% in children with severe scoliosis.

2 | METHODS

This is a cross-sectional retrospective register study based on data from the Cerebral Palsy Follow-Up Programme (CPUP). CPUP is a combined Swedish national quality register and follow-up programme that systematically follows individuals with CP over time.[14] Neuropaediatricians confirm or rule out the CP diagnosis according to the guidelines and definitions set by the *Surveillance of Cerebral Palsy Network in Europe* [15] by age 4 years. Assessment schedules based on GMFCS level and age are used for physiotherapy, occupational therapy and radiographic monitoring of hip displacement. Children at GMFCS level I are examined by their physiotherapists (PT) annually up to 6 years of age and then every second year, whereas children at GMFCS levels II-V are examined twice a year up to 6 years, then once a year. The spines are assessed in seated positions, upright and at forward bending. Any spinal deviations are graded as mild, moderate or severe scoliosis according to the grading scheme presented in Table 1.

TABLE 1 Classification of scoliosis in CPUP

Grade of scoliosis	Description
No scoliosis	No curve
Mild scoliosis	A discrete curve visible only during forward bending
Moderate scoliosis	An obvious curve visible during both extended and forward bending
Severe scoliosis	A pronounced curve preventing the child from attaining an upright position without external support

The standardised clinical spinal assessment in CPUP has shown high interrater reliability, sensitivity, specificity and criterion-related validity compared to radiographic Cobb angle measurements.[16] In addition to physical examinations, the therapists fill out a general survey that includes data on a number of variables, including items on pain. CPUP data from the most recent visits of all children born between 2000-01-01 and 2016-12-31 reported to the registry in 2017-2018 were included.

Age was calculated based on the date of birth and last visit date and was recorded as a continuous variable in years. *Sex* was recorded as a dichotomous variable. The extended and revised version of the GMFCS[17] was used by the PTs to classify *gross motor function* (GMFCS I-V). The screening item on pain ('Does the child, or the parent, state that the child is in pain?') was either self- or, as needed, proxy-reported by the caregiver. If pain were reported, follow-up items on pain site and pain intensity (mild, moderate or severe) were queried. *Pain* was dichotomised as 'present' or 'non-present', both in general (pain at any site) and for back pain specifically. For the purpose of this study, 'neck' pain was included in the 'back' pain category in order to include the entire spine. Furthermore, children with bilateral CP and at least 50% difference in abduction, internal and/or external rotation between the left and right hips were defined as windswept, in accordance with the modified definition of that developed by Young et al.[18] Children who did not have bilateral CP were coded as 'not windswept'. Reduced hip extension was defined as less than 0-degree extension.

2.1 | Statistical analysis

Raw numbers and percentages were calculated on all variables. Chi-square was used to compare differences between groups. Logistic regression was used to regress age, sex, GMFCS level, windswept, and reduced hip extension and self- vs proxy reporting on the presence of back pain. Children who had undergone selective dorsal rhizotomy (n = 80) or used intrathecal baclofen pumps (n = 104) were excluded in the logistic regression analysis. We used 95% confidence intervals (95% CIs) to assess statistical significance.

TABLE 2 Characteristics of the study sample

Characteristics	Boys (n = 2202)	Girls (n = 1581)	Total (N = 3783)
Age, mean (SD)	9.8 (4.36)	10.1 (4.49)	9.95 (4.42)
GMFCS level			
I	980 (45%)	705 (45%)	1685 (45%)
II	329 (15%)	243 (15%)	572 (15%)
III	215 (10%)	139 (9%)	354 (9%)
IV	316 (14%)	240 (15%)	556 (15%)
V	362 (16%)	254 (16%)	616 (16%)
Scoliosis			
No[a]	1618 (73%)	1127 (71%)	2745 (73%)
Mild[a]	355 (16%)	224 (14%)	579 (15%)
Moderate[a]	102 (5%)	88 (6%)	190 (5%)
Severe[a]	58 (3%)	61 (4%)	119 (3%)
Not graded	15 (1%)	17 (1%)	32 (1%)
Scoliosis operated	54 (2%)	64 (4%)	118 (3%)
Windswept	158 (7%)	121 (8%)	279 (7%)
Reduced hip extension[b]	213 (10%)	129 (8%)	342 (9%)
Pain anywhere	845 (38%)	694 (44%)	1539 (41%)
Back pain	114 (5%)	112 (7%)	226 (6%)
Mild	62 (3%)	60 (4%)	122 (3%)
Moderate	42 (2%)	37 (2%)	79 (2%)
Severe	10 (0.5%)	15 (1%)	25 (0.5%)

[a]Children operated for scoliosis not included.
[b]<0 degrees.

The study was approved by the Ethics Board at Lund University (LU-433-99).

3 | RESULTS

In total, 3783 children 1-18 years of age (median 10, SD 4.4 years) were included in the study. The characteristics of the study sample are presented in Table 2.

General pain was reported in 1539 children (40.7%): 694 girls (43.9%) and 845 boys (38.3%) ($P < 0.001$). The pain items were answered by the child in 1718 reports (45.4%), by proxy in 1943 reports (51.3%) and information was missing in 122 reports (3.2%) (Table 3). As expected, the proportion of proxy reports (missing reports excluded) increased with severity of gross motor impairment: GMFCS I (33%), II (54%), III (58%), IV (68%) and V (93%). At each GMFCS level, children who self-reported were more likely to report pain than were those who proxy-reported (Table 3).

Back pain was reported in 226 (6.0%) children. The proportion was higher among girls (7.1%) than boys (5.2%) ($P = 0.017$). The proportion of reported back pain increased with age from <4% before 12 years of age to >12% from 16 years of age (Figure 1).

Scoliosis was reported in 920 children and of these 118 had been operated for scoliosis. The proportion of children with scoliosis increased with GMFCS level (level I = 13.5%, II = 18.5%, III = 24.3%, IV = 35.1%, V = 49.1%). Of the 920 children with scoliosis, 88 (9.6%) reported back pain. Frequency of reported back pain increased with degree of scoliosis, from 4.4% in those who did not have scoliosis to 16.0% in those with severe scoliosis (Figure 2). Pain intensity also increased with degree of scoliosis; moderate back pain increased from 1.5% in children without scoliosis to 6.7% in children with severe scoliosis and severe back pain increased from 0.5% to 3.4% (Figure 2). Of the 118 children who had undergone scoliosis surgery, 15 (12.7%) reported back pain: six mild, eight moderate and one severe back pain.

Results from the logistic regression showed that the odds of reporting back pain increased with age, in girls, in children at GMFCS levels IV-V and in children with scoliosis. Windswept position, reduced hip extension and self-reporting were not statistically significantly associated with back pain (Table 4).

4 | DISCUSSION

In this study, general and back pain were studied in relation to presence and severity of scoliosis, sex, GMFCS level, age, self- vs proxy reporting, windswept and reduced hip extension. Pain intensity in relation to degree of scoliosis was also explored. The overall frequency of reported pain (40.7%) was higher than in a previous Swedish study that included children aged 2-14 years (34%).[11] Pain increases with age in individuals with CP, and the difference can likely be explained by the inclusion of older children in the present study.

The individuals who were able to self-report reported pain more frequently than those who proxy-reported pain for someone else. However, in the regression analysis the OR (1.3) for self-reporting pain was not statistically significant. Although support can be found for proxies both over- and underreporting pain,[13,19,20] it is possible that those unable to communicate are indeed in pain but that the pain is not identified. As pain is a subjective experience, it would be interesting to, in a future study, ask both the child and the parent report on pain and compare the responses. That was not possible to do in the current study. The higher frequency of pain among girls is in agreement with previous studies.[21,22]

Back pain was reported in 15% of all children reporting pain. Previous studies[11,19,22] have shown that pain sites in children with CP

TABLE 3 Pain frequency in children self- versus proxy-reported by gross motor function classification system level

GMFCS level	Pain proxy-reported (%)	Pain self-reported (%)
I	32.7	41.4
II	38.1	49.0
III	31.3	49.0
IV	40.7	43.7
V	52.4	59.5

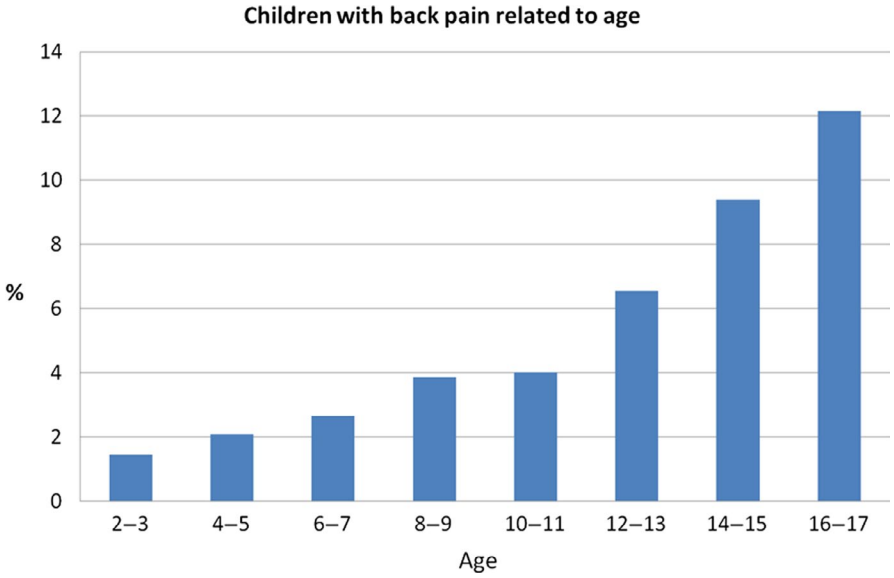

FIGURE 1 Proportion of children and adolescents with cerebral palsy reporting back pain related to age. Six 1-year-old children and 144 18-year-old children were not included in the figure

were primarily located in the lower extremities. Back pain increased significantly with age and was reported in more than 12% of all adolescents 16 years and older. This development raises a concern regarding the magnitude of pain in adults with CP. CPUP continues to follow the total population of individuals with CP in Sweden born 2000 and later, including the further development of pain and scoliosis. It should be noted that back pain in the population of typically developing children is frequent. A meta-analysis reported the mean point prevalence to 12% (95% CI 9-16%).[23] However, it is likely that the characteristics of the pain differ in those with CP.

Scoliosis was a significant risk factor for back pain. Both pain frequency and pain intensity increased with degree of scoliosis. Pain was prevalent even in the children who had undergone surgery for scoliosis. All children operated for scoliosis had severe scoliosis preoperatively, and the pain incidence was only marginally lower than for the children with severe scoliosis who did not have scoliosis surgery. It is possible, however, that the pain severity had decreased after surgery. To analyse the causes of pain in the children who had undergone scoliosis surgery is not possible with registry data. That would require a separate study, which is urgent and planned. Several of the children who had undergone surgery for their scoliosis had persistent scoliosis postoperatively.

Windswept position and reduced hip extension were not associated with back pain in the regression analysis. Children at GMFCS IV and V reported more back pain. These children have most difficulties to change their position in sitting and lying, which can be one reason why they have more back pain.

There were several limitations to our study. Because this was a register-based cross-sectional study, it was not possible to perform in-depth analysis of the causes of pain. For example, we cannot comment on why such a large proportion of the children who had had their scoliosis surgically treated had back pain. It is possible that some of the back pain reported may have been postoperative pain. However, in most cases where the participants have recently

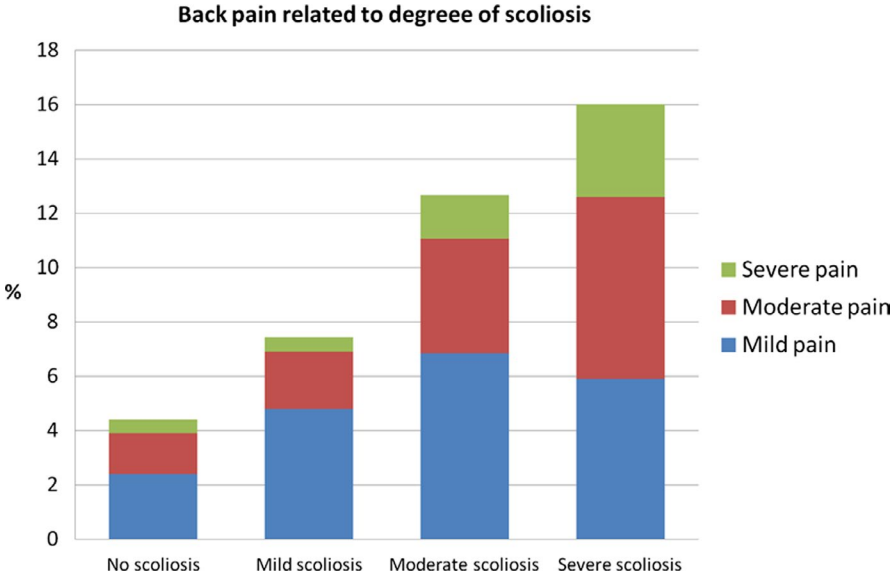

FIGURE 2 Proportion of children and adolescents with cerebral palsy reporting back pain related to degree of scoliosis. Children who underwent scoliosis surgery were excluded

TABLE 4 Logistic regression with the presence of back pain as dependent variable

Variable	Odds ratio	95% CI	
Age[a]	1.18	1.15	1.25
Sex[b]	1.38	1.02	1.89
Scoliosis[c]	1.60*	1.13	2.25
GMFCS[d]			
II	1.36	0.85	2.16
III	1.71	0.99	2.96
IV	1.68	1.03	2.73
V	2.01	1.12	3.63
Windswept[e]	1.37	0.78	2.40
Reduced hip extension[f]	1.27	0.78	2.08
Self-report[g]	1.30	0.87	1.94

Abbreviation: GMFCS, gross motor function classification system.
[a]age stated as increase with 1 year.
[b]Boys as reference.
[c]No scoliosis as reference.
[d]GMFCS I as reference.
[e]No Windswept as reference.
[f]Hip extension > 0 degrees as reference.
[g]Proxy report as reference.

had surgery, the CPUP assessments are postponed to a later date. There were also some missing data regarding proxy versus self-reporting (3.2%) and grading of scoliosis (1.0%). As the proportions were low, they probably did not affect the interpretation of the results in the analysis. There are numerous reasons for why a decision is made to rely on proxy report as opposed to self-report such as young age and intellectual disabilities, and the reason for proxy reporting is not registered in the database. The pain items included in CPUP are screening items. It would have been preferable to have used psychometrically evaluated pain scales. However, in many cases these types of pain scales are rather long and it is not always practically possible to include these at a population base. Pain was not analysed in relation to CP subtype, because there are a lot of missing data regarding CP subtype due to lack of neuropaediatricians in the country.

The main strength of the study is the inclusion of the total population of children and adolescents with CP – also children who cannot report themselves if they have pain.

It is well known that scoliosis can cause a number of secondary problems. This study shows that children with CP and scoliosis more often have back pain and that pain intensity increases with the degree of scoliosis. Many of these individuals cannot explain themselves that they are in pain. It is important that we work proactively to prevent the onset and progression of scoliosis, and that we are aware of analysing whether the scoliosis is painful.

In summary, general pain was reported in 44% of all children, 1-18 years old with CP, and general pain increased with age and was more frequent among girls. Reported frequency of back pain increased with age and degree of scoliosis, from 4% without scoliosis to 16% with severe scoliosis. Intensity of back pain also increased with degree of scoliosis.

ACKNOWLEDGEMENT

This work was supported by Nordforsk grant number 82866; Forte and the Swedish Research Council grant number 218-01468; and Linnea and Josef Carlsson Foundation, Norrbacka Eugenia-stiftelsen, Jerringfonden and Stiftelsen för bistånd åt rörelsehindrade i Skåne. The findings and conclusions in this report are those of the authors and do not necessarily represent the official position of the funders.

CONFLICT OF INTEREST

The authors do not report any competing interests.

ORCID

Gunnar Hägglund https://orcid.org/0000-0002-9593-4927

REFERENCES

1. Rosenbaum P, Paneth N, Leviton A, et al. A report: the definition and classification of cerebral palsy. *Dev Med Child Neurol*. 2007;49:8-14.
2. Wood E, Rosenbaum P. The Gross Motor Function Classification System for cerebral palsy: a study of reliability and stability over time. *Dev Med Child Neurol*. 2000;42:292-296.
3. Alriksson-Schmidt AI, Nordmark E, Czuba T, Westbom L. Stability of the Gross Motor Function Classification System in children and adolescents with cerebral palsy: a retrospective cohort registry study. *Dev Med Child Neurol*. 2017;59:641-646.
4. Palisano RJ, Avery L, Gorter JW, Galuppi B, Westcott McCoy S. Stability of the gross motor function classification system, manual ability classification system, and communication function classification system. *Dev Med Child Neurol*. 2018;60:1026-1030.
5. Hägglund G, Pettersson K, Czuba T, Persson-Bunke M, Rodby-Bousquet E. Incidence of scoliosis in cerebral palsy. *Acta Orthop*. 2018;89:443-447.
6. Koop SE. Scoliosis in cerebral palsy. *Dev Med Child Neurol*. 2009;51:92-98.
7. Bertoncelli CM, Solla F, Loughenbury PR, Tsirikos AI, Bertoncelli D, Rampal V. Risk factors for developing scoliosis in cerebral palsy: a cross-sectional descriptive study. *J Child Neurol*. 2017;32:657-662.
8. Cloake T, Gardner A. The management of scoliosis in children with cerebral palsy: a review. *J Spine Surg*. 2016;2:299-309.
9. Bertoncelli CM, Bertoncelli D, Elbaum L, et al. Validation of a clinical prediction model for the development of neuromuscular scoliosis: a multinational study. *Pediatr Neurol*. 2018;79:14-20.
10. Toovey R, Harvey A, Johnson M, Barker L, Williams K. Outcomes after scoliosis surgery for children with cerebral palsy: a systematic review. *Dev Med Child Neurol*. 2017;59:690-698.
11. Alriksson-Schmidt A, Hägglund G. Pain in children with cerebral palsy. A population based Registry Study. *Acta Paediatr*. 2016;105:665-670.
12. Ramstad K, Jahnsen R, Skjeldal OH, Diseth TH. Parentreported participation in children with cerebral palsy: the contribution of

recurrent musculoskeletal pain and child mental health problems. *Dev Med Child Neurol*. 2012;54:829-835.
13. Parkinson KN, Dickinson HO, Arnaud C, Alan Lyons A, Colver A. SPARCLE group. Pain in young people aged 13–17 years with cerebral palsy: cross-sectional, multicentre European study. *Arch Dis Child*. 2013;98:434-440.
14. Alriksson-Schmidt AI, Arner M, Westbom L, et al. A combined surveillance program and quality register improves management of childhood disability. *Disabil Rehabil*. 2017;39(8):830-836.
15. Surveillance of Cerebral Palsy in Europe. Prevalence and characteristics of children with cerebral palsy in Europe. *Dev Med Child Neurol*. 2002;44:14.
16. Persson-Bunke M, Czuba T, Hägglund G, Rodby-Bousquet E. Psychometric evaluation of spinal assessment methods to screen for scoliosis in children and adolescents with cerebral palsy. *BMC Musculoskelet Disord*. 2015;16:351.
17. Palisano R, Rosenbaum P, Bartlett D, Livingston M. Content validity of the expanded and revised gross motor function classification system. *Dev Med Child Neurol*. 2008;50:744-750.
18. Young NL, Wright JG, Lam TP, Rajaratnam K, Stephens D, Wedge JH. Windswept hip deformity in spastic quadriplegic cerebral palsy. *Pediatr Phys Ther*. 1998;10:94-100.
19. Penner M, Xie WY, Binepal N, Switzer L, Fehlings D. Characteristics of pain in children and youth with cerebral palsy. *Pediatrics*. 2013;132:e407.
20. Hadden KL, LeFort S, O'Brien M, Coyote PC, Guerriere DN. A comparison of observers' and self-report pain ratings for children with cerebral palsy. *J Dev Behav Pediatr*. 2015;36:14-23.
21. Rethlefsen SA, Nguyen DT, Wren TAL, Milewski MD, Kay RM. Knee pain and patellofemoral symptoms in patients with cerebral palsy. *J Pediatr Orthop*. 2015;35:519-522.
22. Dickinson HO, Parkinson KN, Ravens-Sieberer U, et al. Self-reported quality of life of 8-12-year-old children with cerebral palsy: a cross-sectional European study. *Lancet*. 2007;369:2171-2178.
23. Calvo-Munoz I, Gomez-Conesa A, Sanches-Meca J. Prevalence of low back pain in children and adolescents: a meta-analysis. *BMC Pediatr*. 2013;13:14.

How to cite this article: Hägglund G, Czuba T, Alriksson-Schmidt AI. Back pain is more frequent in girls and in children with scoliosis in the context of cerebral palsy. *Acta Paediatr*. 2019;108:2229–2234. https://doi.org/10.1111/apa.14909