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# Relationship between scoliosis, windswept hips and contractures with pain and asymmetries in sitting and supine in 2450 children with cerebral palsy

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**ABSTRACT**

**Purpose:** This cross-sectional study of 2450 children with cerebral palsy aimed to analyse the prevalence and association of scoliosis, windswept hips, hip and knee contractures.

**Methods:** Logistic regression was used to estimate associations with pain, postural asymmetries, and ability to change position for children at Gross Motor Function Classification System (GMFCS) levels I–V, aged 0–18 years.

**Results:** Most children with a deformity or contracture had postural asymmetries in both sitting and supine positions; 10.5% had scoliosis, 8.7% windswept hips, 6.6% hip flexion and 19.2% knee contractures. Severe postural asymmetries increased the likelihood for scoliosis 9 times, for windswept hips 6 to 9 times, and for hip and knee flexion contractures 7 and 12 times respectively, adjusted for age, sex and GMFCS level. Hip flexion contractures and windswept hips increased the likelihood for pain by 1.5–1.6 times.

**Conclusion:** The likelihood of having scoliosis, windswept hips and flexion contractures in the hips and knees increased if the child had postural asymmetries, and for increased age and higher GMFCS levels. Efforts should focus on preventing postural asymmetries from occurring or progressing, and on increasing the child's ability to change position. Reducing postural asymmetries may also reduce the likelihood of pain.

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Cerebral palsy; hip; knee; pain; posture; scoliosis; sitting position; supine position

➤ **IMPLICATIONS FOR REHABILITATION**

- The risk of having scoliosis, windswept hip deformity and flexion contractures in the hips and knees increased if the child had postural asymmetries in sitting or lying.
- Efforts should focus on preventing or reducing postural asymmetries, and on increasing the child's ability to change position.
- Reducing postural asymmetries may also reduce the risk of pain.

## Introduction

Secondary complications often experienced by children with cerebral palsy include a variety of postural asymmetries [1], deformities [2,3], and pain [4], all of which impact upon their ability to socially and physically participate in everyday life [5]. Such deformities can include scoliosis [6,7], hip dislocations [2], windswept deformity; as well as hip and knee contractures [8]. Furthermore, 30–70% of children with cerebral palsy experience pain [9].

Many children with cerebral palsy have difficulties maintaining a symmetrical posture or independently changing position in sitting or lying, which may result in habitual asymmetric postures, potentially leading to deformities and contractures [1]. Adults with cerebral palsy who are unable to change position in lying also have an increased risk of windswept hips and scoliosis [10]. As deformities are observed in children, it is important to determine which variables are associated with their occurrence so that efforts can focus on reducing the risk of their development and progression.

Our aim was to analyse the prevalence of deformities and contractures of the spine, hips, and knees in children with cerebral palsy and their association with postural asymmetries, ability to change position, and pain, considering differences in sex, age, and gross motor function.

## Materials and methods

### *Children and methods*

This cross-sectional study used data of children aged 0–18 years included in the Swedish National Follow-up Programme for People with Cerebral Palsy (CPUP) registry from 1 January 2017 to 30 June 2018. CPUP includes regular clinical examinations to aid in the prevention of deformities and improve physical function in children with cerebral palsy, and has a coverage of >95% of all

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**Table 1.** Participant characteristics of the 2450 children with cerebral palsy.

| Variable | Category | Frequency | Percent |
|---|---|---|---|
| GMFCS | I | 1026 | 41.9 |
| | II | 404 | 16.5 |
| | III | 271 | 11.0 |
| | IV | 386 | 15.8 |
| | V | 363 | 14.8 |
| Age (years)[a] | 0–3 | 236 | 9.6 |
| | 4–6 | 513 | 20.9 |
| | 7–9 | 506 | 20.7 |
| | 10–12 | 529 | 21.6 |
| | 13–15 | 435 | 17.8 |
| | 16–18 | 231 | 9.4 |
| Sex | Boy | 1460 | 59.6 |
| | Girl | 990 | 40.4 |
| Pain | Yes | 938 | 38.3 |
| Deformity | Scoliosis (moderate, severe, fusion) | 257 | 10.5 |
| | Hip Dislocation | 5 | 0.2 |
| | Windswept hips | 213 | 8.7 |
| | Limited Hip Flexion $<90$ | 25 | 1.0 |
| | Hip Flexion Contracture $\geq 10$ | 162 | 6.6 |
| | Knee Flexion Contracture $\geq 10$ | 470 | 19.2 |

[a]Mean (SD) 9.4 (4.3) years.

children with cerebral palsy in Sweden [11]. Children from 4 years of age have their diagnosis of cerebral palsy confirmed by a neuropaediatrician according to the definition by Rosenbaum et al. [12]. The registry uses the Surveillance of Cerebral Palsy in Europe [13] inclusion and exclusion criteria of cerebral palsy.

In the CPUP registry, each child is reviewed clinically twice a year until their sixth birthday and annually thereafter by their local occupational and physiotherapists [8]. Gross motor function of each child was classified using the expanded and revised version of the Gross Motor Function Classification System (GMFCS) [14]. Children with GMFCS level II in the CPUP registry receive radiographic follow-up of their hips at 2 and 6 years of age, whilst children who are at GMFCS levels III–V are reviewed annually until their 8th birthday [8]. Current pain was self- or proxy reported as yes/no to having pain, in response to the question: “Does the person experience pain?” [15].

Passive range of motion (ROM) of knee extension, hip flexion, hip extension, abduction, and internal- and external rotation of the hips were measured in standardized positions using a goniometer. Knee extension was measured in supine with extended hips, while hip extension was measured in prone with both legs lying beyond the end of the plinth and the pelvis held as level as possible with the plinth. Hip abduction in supine was measured with extended hips while rotation of the hips in prone was measured with flexed knees.

Inability to passively get one or both legs straight (hip- or knee flexion contracture of at least 10°) or flex the hips into 90° was classified as a contracture. Windswept hip deformity was calculated based on passive ROM of abduction, internal and external rotation of the hips with a difference of at least 50% between the left and right side using Persson–Bunke’s method [16]. Hip dislocations were defined as Reimers index with a migration percentage of 100%.

Scoliosis was defined as a moderate or severe curve at clinical examination or having a spinal fusion because of scoliosis [17]. Clinical spinal examination was performed with the child sitting on a plinth in an upright position using the forward bending test. Moderate scoliosis (obvious curve present in both upright and forward bending) and severe scoliosis (pronounced curve that prevents upright position without external support) have been reported to have high interrater reliability and high sensitivity and specificity when compared with a radiographic Cobb angle of $\geq 20°$ [17].

The Posture and Postural Ability Scale (PPAS) was used to assess supine lying and sitting posture in the frontal and sagittal planes. It has good interrater reliability and validity for children with cerebral palsy [18]. The scale ranges from full symmetry (6 points) to total asymmetry (0 points). The position of the head, trunk, pelvis, legs, arms/feet and weight bearing is rated as being in midline/symmetric (1 point) or deviating from midline/asymmetric (0 points). We grouped the asymmetries into “no” asymmetry (6 points), “mild” asymmetry (4–5 points), “moderate” asymmetry (2–3 points), and “severe” asymmetry (0–1 points). Similarly, their ability to change or maintain position was rated according to the levels of PPAS ranging from “Able to move into and out of position independently” (level 7) to “Unplaceable in an aligned position” (level 1). We grouped ability to change position into four categories: “changes position” (level 7; reference category), “moves within” (levels 5–6), “maintains”, (levels 3–4) and “cannot maintain” position (levels 1–2). Children were included in our study if they had data reported on posture in either supine or sitting, and for each of the deformities and contractures outcome variables.

### *Statistical analyses*

Frequencies and percentages, *n* (%) were used to describe categorical data, while continuous data were reported as means with accompanying standard deviations (SDs). Age was grouped into six categories: 0–3, 4–6, 7–9, 10–12, 13–15 and 16–18 years old. Pearson’s $\chi^2$-test and the $\chi^2$-test for trend were used for tests of differences between categorical variables. Adjusted and unadjusted binary logistic regression models were used to estimate the magnitude of the association between risk factors and outcomes, presented as odds ratios (ORs) with accompanying 95% confidence intervals (CIs). The OR for having deformity was estimated using postural asymmetry (four categories), ability to change position (four categories), age (six categories), sex, and GMFCS level (five categories) as independent predictors. Full postural symmetry (PPAS 6 points), ability to change position independently (PPAS level 7), lower age (0–3 years), male sex, and better gross motor function (GMFCS level I) were used as reference categories in the regression models. The OR for pain was estimated using each deformity and limited ROM of hip and knee as independent predictors, adjusted for age, sex, and GMFCS level. IBM SPSS Statistics 26 was used for statistical analyses, with *p*-values $< 0.05$ considered statistically significant.

## Results

### *Participants*

Of the 3296 children registered into the CPUP registry a total 2450 children were included, 990 girls (40.4%) and 1460 boys (59.6%) with a mean (SD) age of 9.4 (4.3) years, excluding those with missing ROM data and missing posture data in either sitting or supine positions ($n = 846$). Just over 40% of the children were classified as GMFCS level I. Pain was reported by 938 children (38.3%) (Table 1).

A drop-out analysis was completed comparing the characteristics of the participants group against the non-participant group (age, sex, GMFCS levels). There was no statistically significant difference in the distribution in GMFCS levels or sex between the participants and the non-participants. However, the mean age of the non-participants was 1 year younger (mean 8.38 years;

**Table 2.** Distribution of deformities by sex, age, and GMFCS level for the 2450 participating children.

| | Scoliosis (257) | | Windswept hips (213) | | Hip contracture ≥10° (162) | | Knee contracture ≥10° (470) | |
|---|---|---|---|---|---|---|---|---|
| Variable (*n*) | *n* | % | *n* | % | *n* | % | *n* | % |
| Sex | | | | | | | | |
| Boy (1460) | 134 | 9.2 | 118 | 8.1 | 98 | 6.7 | 295 | 20.2 |
| Girl (990) | 123 | 12.4 | 95 | 9.6 | 64 | 6.5 | 175 | 17.7 |
| Age Groups | | | | | | | | |
| 0–3 yrs (236) | 7 | 3.0 | 6 | 2.5 | 2 | 0.9 | 11 | 4.7 |
| 4–6 yrs (513) | 25 | 4.9 | 24 | 4.7 | 14 | 2.7 | 49 | 9.6 |
| 7–9 yrs (506) | 48 | 9.5 | 32 | 6.3 | 25 | 4.9 | 75 | 14.8 |
| 10–12 yrs (529) | 63 | 11.9 | 51 | 9.6 | 39 | 7.4 | 123 | 23.3 |
| 13–15 yrs (435) | 73 | 16.8 | 61 | 14.0 | 52 | 12.0 | 140 | 32.2 |
| 16–18 yrs (231) | 41 | 17.7 | 39 | 16.9 | 30 | 13.0 | 72 | 31.2 |
| GMFCS level | | | | | | | | |
| GMFCS I (1026) | 15 | 1.5 | 40 | 3.9 | 21 | 2.1 | 22 | 2.1 |
| GMFCS II (404) | 12 | 3.0 | 22 | 5.5 | 19 | 4.7 | 26 | 6.4 |
| GMFCS III (271) | 20 | 7.4 | 27 | 10.0 | 19 | 7.0 | 69 | 25.5 |
| GMFCS IV (386) | 65 | 16.8 | 37 | 9.6 | 43 | 11.1 | 177 | 45.9 |
| GMFCS V (363) | 145 | 39.9 | 87 | 24.0 | 60 | 16.5 | 176 | 48.5 |

25th–75th percentile 4–12 years) than the mean age of the participants (mean 9.35 years; 25th–75th percentile 6–13 years).

### *Deformities, contractures and hip dislocations*

In total 257 children (10.5%) had scoliosis and 213 (8.7%) had windswept hips (Table 1). Knee flexion contractures of ≥10° were found in 470 children (19.2%), while 162 children (6.6%) had hip flexion contractures. Only 5 children (0.2%) had dislocated hips and 25 children (1%) had <90° of hip flexion (Table 1). As there were too few observations in these two sub-groups to perform any meaningful statistical analyses, hip dislocation and hip flexion limitations were omitted from further analyses.

All deformities and contractures were noted across both sexes, all age groups, and all GMFCS levels, although most frequently in the 16–18 years age group, GMFCS level V, and for children with moderate or severe postural asymmetries (Table 2).

More children unable to maintain position in either supine or sitting positions had scoliosis, windswept hips, hip or knee flexion contractures. A higher proportion of these children also had severe postural asymmetries in both sitting and supine positions (Table 3).

An asymmetric sitting posture increased the likelihood of scoliosis (OR 9.1, CI 4.9–16.9), and windswept hips (OR 5.7, CI 3.2–9.8), and an asymmetric supine posture increased the likelihood of windswept hips (OR 8.8, CI 4.9–15.8), hip flexion contractures (OR 6.7, CI 3.4–13.1), and knee flexion contractures (OR 12.2, CI 7.1–20.8) when adjusted for age, sex, and GMFCS level. Additionally, the likelihood of having scoliosis was higher in girls (OR 1.7, CI 1.2–2.3) (Table 4).

### *Pain*

A higher proportion of the children with windswept hips (53%), hip contracture (52%) and scoliosis (51%) had pain, compared to children with knee contracture (44%). However, only windswept hips (OR 1.6, CI 1.2–2.2) and hip flexion contractures (OR 1.5, CI 1.1–2.1) were significantly associated with pain when adjusted for age, sex, and GMFCS level (Table 5).

## Discussion

This study found that children with postural asymmetries in sitting or in supine had an increased likelihood of having scoliosis, windswept hip deformities, and contractures of the hips and knees. This likelihood increased as the severity of postural asymmetries increased. Children unable to change position in sitting or lying were more likely to have deformities and contractures. These results are consistent with previous studies and add to Fulford and Brown's [19] seminal work with children and Rodby-Bousquet et al.'s [20] work with young adults showing that immobility and asymmetric positions sustained for longer periods of time may result in deformities in children and adults with cerebral palsy. Similarly, Ágústsson et al. [10] reported that adults with cerebral palsy who habitually lay in an asymmetric posture and were unable to change position had a higher likelihood of having scoliosis and windswept hip deformities.

Knee flexion contractures (19.2%) were most frequent, with an incidence similar to that of Cloodt et al. [21], who reported that one in four children aged ≤15 years had knee contractures. In a later study, Cloodt et al. [8] explored the location of first lower limb contractures in children with cerebral palsy and found that knee contractures were the first lower limb contracture to occur in children with GMFCS levels III-V.

Scoliosis (10.5%) was the second most common deformity in our study and occurred more frequently in girls than in boys. The prevalence of scoliosis in children is comparable to Hägglund et al. [6] who found a prevalence of 15%, although their cohort of children followed from birth to 25 years of age only included the Southern Sweden region. Likewise, Persson-Bunke et al. [22] reported a prevalence of 11% in a total population of Swedish children aged 4–18 years. Pettersson et al. [23] reported a higher prevalence of moderate to severe scoliosis in girls. Additionally, Pettersson et al. [7] identified that being a girl and having knee flexion contracture were significant predictors for the development of severe scoliosis. Therefore, girls and children at GMFCS IV and V with knee flexion contractures should be closely monitored in efforts to identify children at risk early and allow timely interventions to help prevent both occurrence and progression of scoliosis. Furthermore, we found that postural asymmetries in sitting and supine is associated with scoliosis. This might be explained by the Hueter–Volkmann principle where asymmetric loading of the vertebrae can lead to asymmetric growth and structural spinal deformities [24].

We identified 5 children (0.2%) with hip dislocations and 213 (8.7%) with windswept deformity. This is similar to Hägglund et al. [3] using an earlier cohort of 214 children, in which only one child had hip dislocation and 9% windswept hip deformity. For adults aged 16–73 years old, Ágústsson et al. [10] found that having scoliosis and windswept hips was associated with spending

**Table 3.** Distribution of deformities and contractures by pain, postural ability and postural asymmetry for the 2450 participating children.

| | Scoliosis ($n = 257$) | | Windswept hips ($n = 213$) | | Hip contracture $\geq 10°$ ($n = 162$) | | Knee contracture $\geq 10°$ ($n = 470$) | |
|---|---|---|---|---|---|---|---|---|
| Variable (*n*) | *n* | % | *n* | % | *n* | % | *n* | % |
| **Pain** | | | | | | | | |
| Yes (938) | 131 | 14.0 | 113 | 12.1 | 85 | 9.1 | 207 | 22.1 |
| **Postural ability** | | | | | | | | |
| *Supine ability* | | | | | | | | |
| Changes position (1720) | 58 | 3.4 | 82 | 4.8 | 64 | 3.7 | 151 | 8.8 |
| Moves within position (357) | 59 | 16.5 | 39 | 10.9 | 35 | 9.8 | 139 | 38.9 |
| Maintains position (285) | 103 | 36.1 | 65 | 22.8 | 37 | 13.0 | 132 | 46.3 |
| Cannot maintain position (80) | 37 | 46.3 | 26 | 32.5 | 26 | 32.5 | 47 | 58.8 |
| *Sitting ability* | | | | | | | | |
| Changes position (1564) | 41 | 2.6 | 74 | 4.7 | 53 | 3.4 | 97 | 6.2 |
| Moves within position (219) | 27 | 12.3 | 15 | 6.9 | 21 | 9.6 | 83 | 37.9 |
| Maintains position (149) | 30 | 20.1 | 20 | 13.4 | 16 | 10.7 | 70 | 47.0 |
| Cannot maintain position (501) | 156 | 31.1 | 101 | 20.2 | 69 | 13.8 | 218 | 43.5 |
| **Postural asymmetry** | | | | | | | | |
| *Supine asymmetry frontal* | | | | | | | | |
| No (1371) | 27 | 2.0 | 46 | 3.4 | 27 | 2.0 | 57 | 4.2 |
| Mild (557) | 53 | 9.5 | 40 | 7.2 | 42 | 7.5 | 134 | 24.1 |
| Moderate (305) | 81 | 26.6 | 54 | 17.7 | 45 | 14.8 | 141 | 46.2 |
| Severe (217) | 96 | 44.2 | 73 | 33.6 | 48 | 22.1 | 138 | 63.6 |
| *Supine asymmetry sagittal* | | | | | | | | |
| No (1314) | 32 | 2.4 | 46 | 3.5 | 26 | 2.0 | 35 | 2.7 |
| Mild (614) | 70 | 11.4 | 47 | 7.7 | 38 | 6.2 | 145 | 23.6 |
| Moderate (332) | 82 | 24.7 | 59 | 17.8 | 52 | 15.7 | 171 | 51.5 |
| Severe (190) | 73 | 38.4 | 61 | 32.1 | 46 | 24.2 | 119 | 62.6 |
| *Sitting asymmetry frontal* | | | | | | | | |
| No (1267) | 20 | 1.6 | 46 | 3.6 | 27 | 2.1 | 75 | 5.9 |
| Mild (595) | 54 | 9.1 | 40 | 6.7 | 46 | 7.7 | 131 | 22.0 |
| Moderate (351) | 75 | 21.4 | 56 | 16.0 | 40 | 11.4 | 126 | 35.9 |
| Severe (237) | 108 | 45.6 | 71 | 30.0 | 49 | 20.7 | 138 | 58.2 |
| *Sitting asymmetry sagittal* | | | | | | | | |
| No (1193) | 27 | 2.3 | 44 | 3.7 | 32 | 2.7 | 70 | 5.9 |
| Mild (600) | 66 | 11.0 | 58 | 9.7 | 47 | 7.8 | 130 | 21.7 |
| Moderate (523) | 121 | 23.1 | 71 | 13.6 | 56 | 10.7 | 197 | 37.7 |
| Severe (134) | 43 | 32.1 | 40 | 29.9 | 27 | 20.1 | 73 | 54.5 |

**Table 4.** Logistic regression analysis with adjusted odds ratios (ORs) and 95% confidence intervals (CIs) for deformities and contractures.

| | Scoliosis (Sitting Frontal) | | | | Windswept hips (Sitting Frontal) | | | | Windswept hips (Supine Frontal) | | | | Hip flexion contracture (Supine Sagittal) | | | | Knee flexion contracture (Supine Sagittal) | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | OR | 95% CI | | *p*-value | OR | 95% CI | | *p*-value | OR | 95% CI | | *p*-value | OR | 95% CI | | *p*-value | OR | 95% CI | | *p*-value |
| Sex | | | | | | | | | | | | | | | | | | | | |
| Boy | *Ref.* | | | | *Ref.* | | | | *Ref.* | | | | *Ref.* | | | | *Ref.* | | | |
| Girl | 1.7 | 1.2 | 2.3 | 0.001 | 1.2 | 0.9 | 1.7 | 0.17 | 1.3 | 1.0 | 1.7 | 0.12 | 1.0 | 0.7 | 1.4 | 0.93 | 0.8 | 0.6 | 1.1 | 0.15 |
| Age group | | | | | | | | | | | | | | | | | | | | |
| 0–3 yrs | *Ref.* | | | | *Ref.* | | | | *Ref.* | | | | *Ref.* | | | | *Ref.* | | | |
| 4–6 yrs | 2.2 | 0.9 | 5.4 | 0.09 | 2.2 | 0.9 | 5.4 | 0.11 | 2.2 | 0.9 | 5.7 | 0.09 | 4.0 | 0.9 | 17.7 | 0.07 | 3.2 | 1.6 | 6.7 | 0.001 |
| 7–9 yrs | 5.0 | 2.1 | 11.9 | <0.001 | 2.8 | 1.1 | 7.0 | 0.03 | 2.8 | 1.1 | 6.9 | 0.03 | 6.6 | 1.5 | 28.5 | 0.01 | 5.2 | 2.6 | 10.6 | <0.001 |
| 10–12 yrs | 6.9 | 2.9 | 16.1 | <0.001 | 4.5 | 1.9 | 10.9 | 0.001 | 4.3 | 1.8 | 10.3 | 0.001 | 9.7 | 2.3 | 40.9 | 0.002 | 10.7 | 5.3 | 21.3 | <0.001 |
| 13–15 yrs | 10.7 | 4.5 | 25.1 | <0.001 | 6.8 | 2.8 | 16.3 | <0.001 | 6.1 | 2.5 | 14.8 | <0.001 | 15.6 | 3.7 | 65.5 | <0.001 | 19.6 | 9.7 | 39.7 | <0.001 |
| 16–18 yrs | 13.5 | 5.5 | 33.2 | <0.001 | 8.7 | 3.5 | 21.6 | <0.001 | 8.0 | 3.2 | 20.0 | <0.001 | 18.1 | 4.2 | 78.1 | <0.001 | 19.4 | 9.1 | 41.1 | <0.001 |
| GMFCS level | | | | | | | | | | | | | | | | | | | | |
| GMFCS I | *Ref.* | | | | *Ref.* | | | | *Ref.* | | | | *Ref.* | | | | *Ref.* | | | |
| GMFCS II | 1.5 | 0.7 | 3.3 | 0.31 | 1.1 | 0.7 | 2.0 | 0.66 | 1.1 | 0.6 | 1.9 | 0.80 | 1.6 | 0.8 | 3.2 | 0.14 | 1.8 | 1.0 | 3.3 | 0.07 |
| GMFCS III | 3.1 | 1.5 | 6.4 | 0.002 | 1.8 | 1.1 | 3.2 | 0.03 | 1.6 | 0.9 | 2.8 | 0.10 | 2.0 | 1.0 | 4.0 | 0.06 | 8.5 | 4.9 | 14.8 | <0.001 |
| GMFCS IV | 5.5 | 2.9 | 10.5 | <0.001 | 1.2 | 0.7 | 2.0 | 0.56 | 0.9 | 0.5 | 1.6 | 0.78 | 2.2 | 1.1 | 4.2 | 0.02 | 14.5 | 8.5 | 24.5 | <0.001 |
| GMFCS V | 16.4 | 8.6 | 31.5 | <0.001 | 2.8 | 1.7 | 4.9 | <0.001 | 2.0 | 1.1 | 3.5 | 0.02 | 2.9 | 1.5 | 5.7 | 0.002 | 13.5 | 7.7 | 23.6 | <0.001 |
| Asymmetry | | | | | | | | | | | | | | | | | | | | |
| No (6 p) | *Ref.* | | | | *Ref.* | | | | *Ref.* | | | | *Ref.* | | | | *Ref.* | | | |
| Mild (4–5 p) | 3.1 | 1.8 | 5.6 | <0.001 | 1.6 | 1.0 | 2.6 | 0.05 | 2.0 | 1.2 | 3.3 | 0.004 | 2.2 | 1.2 | 3.9 | 0.007 | 4.7 | 3.1 | 7.2 | <0.001 |
| Moderate (2–3 p) | 5.0 | 2.8 | 8.9 | <0.001 | 3.4 | 2.1 | 5.6 | <0.001 | 4.8 | 2.9 | 8.2 | <0.001 | 4.9 | 2.7 | 9.1 | <0.001 | 10.8 | 6.8 | 17.2 | <0.001 |
| Severe (0–1 p) | 9.1 | 4.9 | 16.9 | <0.001 | 5.7 | 3.2 | 9.8 | <0.001 | 8.8 | 4.9 | 15.8 | <0.001 | 6.7 | 3.4 | 13.1 | <0.001 | 12.2 | 7.1 | 20.8 | <0.001 |

Posture and Postural Ability Scale (PPAS) no asymmetry in sitting or supine, Gross Motor Function Classification System (GMFCS) level I, youngest age group (0–3 years), and being a boy were used as reference categories. Age was used as a categorical variable. All variables were adjusted for all other variables in the model.

>8 h lying in the same position and having the inability to change position. Although they reported a higher incidence of both deformities, their older participants had not been followed within a surveillance program as children, so our lower numbers either serve to illustrate the benefits of regular and early surveillance or these deformities are formed later in life.

**Table 5.** Logistic regression analysis with odds ratios (ORs) and 95% confidence intervals (CIs) for pain with each individual deformity unadjusted and adjusted for GMFCS level, age and sex.

| | Unadjusted | | | | Adjusted | | | |
|---|---|---|---|---|---|---|---|---|
| Deformity | OR | 95% CI | | *p*-value | OR | 95% CI | | *p*-value |
| Scoliosis | 1.9 | 1.4 | 2.4 | <0.001 | 1.3 | 1.0 | 1.8 | 0.089 |
| Windswept hips | 2.1 | 1.6 | 2.9 | <0.001 | 1.6 | 1.2 | 2.2 | 0.002 |
| Hip contracture $\geq 10^\circ$ | 2.0 | 1.4 | 2.8 | <0.001 | 1.5 | 1.1 | 2.1 | 0.025 |
| Knee contracture $\geq 10^\circ$ | 1.4 | 1.2 | 1.8 | 0.001 | 1.0 | 0.8 | 1.3 | 0.893 |

Reference categories: Gross Motor Function Classification System (GMFCS) level I; boys; 0–3 years. Age was used as a categorical variable.

Few children were unable to flex their hips to 90° (1%), and so this low prevalence did not allow further analyses. However, in a study of adults with cerebral palsy Ágústsson et al. [16] reported a prevalence of 9%, and an increased risk of scoliosis and windswept hip deformity for those with asymmetric limited hip flexion.

Rodby-Bousquet et al. [20] recognised that for young adults with cerebral palsy there was a high association between having hip flexion contracture and postural asymmetry. In our study, 6.6% children had hip flexion contractures of >10°, and the odds of having a hip flexion contracture was 6.7 times higher if the child had postural asymmetries. Our finding that deformities and contractures are already established across all age groups of children is important as it highlights that children with cerebral palsy are at risk of developing these, but more so if they have postural asymmetries.

It is therefore necessary that we aim to reduce the likelihood of deformities and contractures by focusing attention on careful postural alignment and symmetrical positioning, especially in children unable to change position independently. The possibility that deformities and contractures might become established early in life reinforces the importance of regular surveillance to monitor posture and ROM to provide as early an intervention as necessary.

We found a pain prevalence of 38.3%, which is considerably lower than the 74–77% reported for 13–17 year olds in a European multicentre study by Parkinson et al. [25]. Although lower, our 38.3% is still a high number of children potentially experiencing pain that can impact on their health, wellbeing, and participation in everyday life. A possible explanation for this discrepancy may be the use of different pain assessments, and that we included reports of current pain, whilst Parkinson et al. [25] reported occurrence of pain within the previous week and year.

Eriksson et al. [9] reported that 44% of Swedish children with cerebral palsy aged 4–18 years old had pain, that the pain increased with age, and that it was more frequent in the lower limbs. Our results with 0–18 year olds show a similar incidence of pain, and we found a significant association between pain and deformities/contractures specifically at the hips. Children with windswept hips or hip flexion contractures were more likely to have pain. Our findings indicate a higher likelihood of pain in the hips despite these children presenting with a higher incidence of scoliosis and knee flexion contractures. This association reinforces the importance of interventions to prevent hip deformities/contractures.

Recent work by Casey et al. [1] shows an association between pain and postural asymmetries in children with cerebral palsy, and also that inability to change position increases the likelihood of having postural asymmetries. Our results indicate a clear association between postural asymmetries in sitting and lying with deformities/contractures of the spine, hips, and knees. Therefore, it is vital that interventions are directed at postural asymmetries whilst they are still reducible to inhibit the development of fixed deformities and contractures. One such intervention strategy is to reduce the time spent in asymmetric postures through the provision of appropriate supports, such as individually tailored seating systems, wheelchairs, or orthoses, and finding non-harmful and comfortable sleeping positions to reduce stress and strain on tissues for children unable to independently change or maintain their position in sitting or lying. This in turn will support the prevention of habitual postural asymmetries, tissue adaption, and ultimately scoliosis, windswept hips, and contractures from either occurring or progressing, and potentially prevent these children from experiencing pain as a result.

### *Limitations*

The cross-sectional design means that we can show associations, but not causal relationships between deformities, contractures, and postural asymmetries. We did not investigate the location or severity of pain, only the association between current pain and deformities/contractures of the spine, hips, or knees. However, it is concerning that so many children had pain; and, that we found an association between having pain and having deformities/contractures.

We were unable to include postural ability from the PPAS as an independent variable in the logistic regression analyses due to collinearity with GMFCS. Since GMFCS is recognized as an international standard used to classify cerebral palsy, we opted to retain the GMFCS level in these analyses.

A further consideration is the categorization we used in our methodology. In order to interpret the data for scoliosis, hip and knee flexion contractures we applied cut-off levels for our variables. Specifically, for scoliosis we used the categories of 'none' or 'moderate to severe' and did not include mild curves only visible at forward bending as having scoliosis. This was because the definition of 'mild' scoliosis used in the follow-up program cannot be distinguished from 'no' scoliosis on spinal radiographs in sitting [17]. Similarly, for both hip and knee flexion contractures the prevalence may have been affected as we applied a cut-off level of a loss of hip or knee extension of ≥ 10° to reduce the impact of any measurement error. However, a major strength of this study is that it is based upon a total population of children with cerebral palsy, including children of all ages and GMFCS levels.

## Conclusion

In summary, the occurrence of deformities and contractures of the spine, hips, and knees among children with cerebral palsy are strongly associated with having postural asymmetry, whilst pain was mostly associated with having windswept hip deformity or hip flexion contractures. Whilst this is not a causal relationship, clinical interventions for children with cerebral palsy should address preventing and reducing postural asymmetries to lower the likelihood of these deformities and contractures occurring and subsequently reduce the likelihood for pain.

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## Ethical approval

The Medical Research Ethics Committee at Lund University approved the study (383/2007, 443-99), and permission was obtained to extract data from the CPUP registry. Legal caregivers of all participants consented to research based on the data held in the registry.

## Disclosure statement

The authors declare no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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## Data availability statement

The dataset analysed during the current study is part of the CPUP registry.

## References

[1] Casey J, Rosenblad A, Rodby-Bousquet E. Postural asymmetries, pain, and ability to change position of children with cerebral palsy in sitting and supine: a cross-sectional study. Disabil Rehabil. 2020. DOI:10.1080/09638288.2020.1834628.
[2] Hägglund G, Alriksson-Schmidt A, Lauge-Pedersen H, et al. Prevention of dislocation of the hip in children with cerebral palsy. Bone Joint J. 2014;96-B(11):1546–1552.
[3] Hägglund G, Lauge-Pedersen H, Persson Bunke M, et al. Windswept hip deformity in children with cerebral palsy: a population-based prospective follow-up. J Child Orthop. 2016;10(4):275–279.
[4] Tedroff K, Gyllensvärd M, Löwing K. Prevalence, identification, and interference of pain in young children with cerebral palsy: a population-based study. Disabil Rehabil. 2021;43(9):1292–1297.
[5] Michelsen SI, Flachs EM, Damsgaard MT, et al. European study of frequency of participation of adolescents with and without cerebral palsy. Eur J Paediatr Neurol. 2014;18(3):282–294.
[6] Hägglund G, Pettersson K, Czuba T, et al. Incidence of scoliosis in cerebral palsy. Acta Orthop. 2018;89(4):443–447.
[7] Pettersson K, Wagner P, Rodby-Bousquet E. Development of a risk score for scoliosis in children with cerebral palsy. Acta Orthop. 2020;91(2):203–208.
[8] Cloodt E, Wagner P, Lauge-Pedersen H, et al. Knee and foot contracture occur earliest in children with cerebral palsy: a longitudinal analysis of 2,693 children. Acta Orthop. 2021;92(2):222–227.
[9] Eriksson E, Hagglund G, Alriksson-Schmidt AI. Pain in children and adolescents with cerebral palsy – a cross-sectional register study of 3545 individuals. BMC Neurol. 2020;20(1):15.
[10] Ágústsson A, Sveinsson T, Pope P, et al. Preferred posture in lying and its association with scoliosis and windswept hips in adults with cerebral palsy. Disabil Rehabil. 2019;41(26):3198–3202.
[11] Westbom L, Hagglund G, Nordmark E. Cerebral palsy in a total population of 4-11 year olds in Southern Sweden. Prevalence and distribution according to different CP classification systems. BMC Pediatr. 2007;7:41.
[12] Rosenbaum P, Paneth N, Leviton A, et al. A report: the definition and classification of cerebral palsy april 2006. Dev Med Child Neurol Suppl. 2007;109:8–14.
[13] Surveillance of Cerebral Palsy in Europe. Surveillance of cerebral palsy in Europe (SCPE): a collaboration of cerebral palsy surveys and registers. Devel Med Child Neurol. 2000;42:16–24.
[14] Palisano RJ, Rosenbaum P, Bartlett D, et al. Content validity of the expanded and revised gross motor function classification system. Dev Med Child Neurol. 2008;50(10):744–750.
[15] Westbom L, Rimstedt A, Nordmark E. Assessments of pain in children and adolescents with cerebral palsy: a retrospective population-based registry study. Dev Med Child Neurol. 2017;59(8):858–863.
[16] Ágústsson A, Sveinsson Þ, Rodby-Bousquet E. The effect of asymmetrical limited hip flexion on seating posture, scoliosis and windswept hip distortion. Res Dev Disabil. 2017;71:18–23.
[17] Persson-Bunke M, Czuba T, Hagglund G, et al. Psychometric evaluation of spinal assessment methods to screen for scoliosis in children and adolescents with cerebral palsy. BMC Musculoskelet Disord. 2015;16:351.
[18] Rodby-Bousquet E, Persson-Bunke M, Czuba T. Psychometric evaluation of the posture and postural ability scale for children with cerebral palsy. Clin Rehabil. 2016;30(7):697–704.
[19] Fulford FE, Brown JK. Position as a cause of deformity in children with cerebral palsy. Dev Med Child Neurol. 1976;18(3):305–314.
[20] Rodby-Bousquet E, Czuba T, Hagglund G, et al. Postural asymmetries in young adults with cerebral palsy. Dev Med Child Neurol. 2013;55(11):1009–1015.
[21] Cloodt E, Rosenblad A, Rodby-Bousquet E. Demographic and modifiable factors associated with knee contracture in children with cerebral palsy. Dev Med Child Neurol. 2018;60(4):391–396.
[22] Persson-Bunke M, Hägglund G, Lauge-Pedersen H, et al. Scoliosis in a total population of children born with cerebral palsy. Spine. 2012;37:E708–E13.
[23] Pettersson K, Rodby-Bousquet E. Prevalence and goal attainment with spinal orthoses for children with cerebral palsy. J Pediatr Rehabil Med. 2019;12(2):197–203.
[24] Stokes IA, Spence H, Aronsson DD, et al. Mechanical modulation of vertebral body growth. Implications for scoliosis progression. Spine. 1996;21(10):1162–1167.
[25] Parkinson KN, Dickinson HO, Arnaud C, et al. Pain in young people aged 13 to 17 years with cerebral palsy: cross-sectional, multicentre european study. Arch Dis Child. 2013;98(6):434–440.