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RESEARCH PAPER



Preferred posture in lying and its association with scoliosis and windswept hips in adults with cerebral palsy

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ABSTRACT

Objective: The aim of this study was to clarify the association of scoliosis and windswept hips with immobility, lying position, and time in lying, in adults with cerebral palsy (CP).

Methods: This cross-sectional study included 830 adults (469 males and 361 females) with a diagnosis of CP, 16–73 years, and classified at levels I–V according to the Gross Motor Function Classification System (GMFCS). Subjects' Gross motor function classification system level, presence and severity of scoliosis, hip and knee joint range of movement, lying position, postural ability in lying, and time in lying were used to identify connections between them.

Results: Adults who are immobile in the lying position have higher odds of both scoliosis and windswept hips. Spending more than 8 h daily in the same lying position, increased the odds of having scoliosis, while lying solely in a supine position, resulted in higher odds of windswept hips.

Conclusions: The “preferred” habitual posture frequently observed in immobile adults with CP, leads to established distortion of their body shape. The results indicate the need for early introduction of appropriate posture control, in immobile individuals with CP, from a young age.

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Adult; cerebral palsy; scoliosis; wind-sweeping; hip

► IMPLICATIONS FOR REHABILITATION

- The preferred posture, observed in immobile adults with cerebral palsy, leads to a distortion of their body shape.
- One in four adults with cerebral palsy use only one position when in bed.
- The results indicate the need for early introduction of appropriate posture control in individuals unable to change position.



Introduction

Cerebral palsy (CP) is a neurological disorder, caused by a non-progressive brain injury, in the developing foetal or infant brain [1]. Even though the brain injury is non-progressive, secondary complications are prevalent. Limited mobility and postural asymmetry predispose to, for example, tissue adaptation, scoliosis, pelvic obliquity, hip sub/dislocation, and wind-sweeping particularly in the non-ambulant individual [2]. These complications increase the risk of further problems such as pain, pressure ulcers, respiratory, and urinary tract infections [3].

Preferred posture is used to describe a habitual posture, that is, one where the body returns to its original attitude after correction or change of position [3]. It indicates that the tissues of the body have adapted physiologically (plastic adaption) to a particular posture. It is deemed to be the result of extended periods of time in the same position. In a person with disabilities unable to change position the preferred posture is compounded by gravity, leading to further tissue adaptation. There are indications that these preferred postures are established in unborn babies [4], and maintained after birth [5] in babies unable to change from a lying position.

It is the asymmetries of posture, resulting in unequal forces acting on the tissues, compounded by long periods in one position that are thought to predispose to the problems of wind-sweeping, scoliosis, and other distortions of body structure [3,6,7]. Evidence of an association between the severity of CP, hip dislocation, pelvic obliquity, and scoliosis [8] is weak. Others have shown that a child's preferred lying posture is associated with the direction of wind-sweeping, hip dislocation, and lateral scoliosis curve [2,7]. It has been suggested that prone and side-lying may increase the risk of scoliosis and recommend use of the supine position instead [9]. Rodby-Bousquet et al. found that the time spent in one position predisposes to the development of contractures and distortion [10] due to plastic adaptation of the body structures.

Letts et al. [11] defined a windblown hip syndrome as a triad of scoliosis, pelvic obliquity, and wind-swept hips, without defining windswept hips specifically. Lonstein and Beck [12] defined windswept hips as an abduction contracture in one hip combined with an adduction contracture of the other hip. This definition has been used by other researchers [2,13] until Persson-Bunke et al. [14] refined it by describing windswept hips as a combination of abduction and external rotation of one hip with the

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opposite hip in adduction and internal rotation. The prevalence of windswept hips is 18% in the CP population [15], and will, in its severest cases, lead to hip dislocation [2,11].

In a total population of children with CP in Sweden, the prevalence of scoliosis is 11%, which increases with Gross Motor Function Classification System (GMFCS) level and, most significantly, with age [16]. In young adults, with GMFCS levels IV–V, there is a 50% risk of scoliosis by the age of 18 years. When analysing the development of scoliosis in individuals with CP and windswept hips, it was found that a hip dislocation was followed by pelvic obliquity and then scoliosis in three out of four non-ambulatory individuals [11]. Contracture in the knee joint is frequently reported among adults with CP [17], up to 60% cannot fully extend one or both knees, and is associated with postural asymmetries in both supine and standing position [10].

The aim of this study was to examine the association of scoliosis and windswept hips with immobility, lying position, and time in lying, in all adults, 16 years and older, with CP reported into the Swedish national CP surveillance programme and registry (CPUP) [18]. Immobility was defined as inability to move within or change position. Lying position was defined as their default sleeping and resting position.

Methods

Ethical approval was granted by the Medical Research Ethics Committee at Lund University (LU 2009–341), and permission was obtained to extract data from the Swedish national CP surveillance Programme and registry (CPUP). All participants on the register consent to research based on reported data.

Design

This cross-sectional study was based on data from the CPUP registry in Sweden [18]. Data were extracted from the most recent reports for all adults with CP registered between 1 January 2013 and 31 December 2016. Inclusion and exclusion criteria for the CP-diagnosis were defined by the Surveillance of Cerebral Palsy in Europe [19] with the brain injury occurring before the age of two years. Subtypes were classified as spastic unilateral, spastic bilateral, ataxic, and dyskinetic CP.

Assessment protocol

All assessments were performed in a standardised manner by local physiotherapists and occupational therapists throughout the country. They were trained in using the standardised CPUP assessment form and its accompanying manual. Both are attainable on the CPUP website (<http://cpup.se/in-english/manuals-and-evaluation-forms/>). These assessments are included as part of the national surveillance programme and entered into the CPUP web-based registry. The schedule of assessments is based on age and levels of motor function using the expanded and revised version of the GMFCS, age band 12–18 years [20]. Adults classified at GMFCS III–V are examined every year, GMFCS II every second year, and adults at GMFCS I every third year. Even though GMFCS was developed for children, it has also been shown to be accurate for use in adults with CP [21,22].

Passive joint range of movement for knee extension, hip extension, abduction, and external and internal rotation was measured with a goniometer in a standardised supine position. Lying positions and time in lying were either self-reported by the individual or reported by caregivers who know the respective individuals

very well. Lying positions were defined as sleeping and resting positions over a 24 h cycle as supine, prone, right side lying, or left side-lying with the possibility of choosing 1–4 positions. Time in lying in any one position was reported as less than 8 h, 8–12 h, or more than 12 h, within a 24 h period, with the possibility of choosing just one item.

Scoliosis

In CPUP, scoliosis was rated as no scoliosis, mild, moderate, or severe using a clinical spinal examination with high psychometric properties for neuromuscular scoliosis in individuals with CP [23]. In this study, mild scoliosis (a discrete curve only visible in forward bending) was treated as not having scoliosis. Individuals operated for scoliosis were included in the study. Those reported to have a remaining moderate or severe scoliosis after surgery were included in the scoliosis group, if not, they were treated as not having scoliosis.

Posture and Postural Ability Scale

The preferred posture and postural ability in lying were assessed using the 7-point ordinal scale of the Posture and Postural Ability Scale (PPAS) [24], which has high psychometric properties for adults with CP. Postural ability ranges from level “1 – unplaceable” to level “7 – able to move into and out of position”. PPAS level 1–3 indicates that the individual has little or no ability to counteract gravity or change position and is referred to as immobile in this study. Individuals at level 3 can maintain a position when placed by another person but cannot move. Individuals at level 4–7 have the ability to move within a position and ultimately, change position.

Windswept hips

Windswept hips consist of abduction and external rotation of one hip, with the opposite hip in adduction and internal rotation. The presence of windswept hips was calculated from hip range of movement values using Persson-Bunke’s formula for calculation, not through a clinical analysis of posture [14]. The direction of the windswept hips was confirmed by using a modified version of Porter’s formula [2]. Both formulas are modified versions from Young’s work [13]. Those who were not windswept according to both methods were determined as not windswept. In Persson-Bunke’s method, at least 50% difference was needed between right and left side in either: hip abduction, hip internal, or hip external rotation, to define the presence of windswept hips. Values lower than 0.5 or higher than 2 were considered windswept. In Porter’s modified version, the hip range of movement (hip abduction, hip external rotation, and hip internal rotation) on each side was added together (hip internal rotation, being in opposite direction to hip abduction and external rotation, having a minus value). The left side was then subtracted from the right side. Threshold value was 20°, plus value of 20° or higher indicated windswept over to the right, and a minus value of –20° or lower indicated windswept to the left.

Statistical analyses

Spearman’s correlation coefficients (r_s) were calculated for the association between GMFCS levels on one hand, and scoliosis and windswept hips on the other. The interpretation of the strength of the Spearman’s correlation coefficient was: $r_s > 0.70$ strong

relationship; $r_s = 0.70$ – 0.30 moderate relationship; and $r_s < 0.30$ weak relationship. Logistic regression analysis was used to investigate the association between variables. Scoliosis and windswept hips were used as outcome variables. Explanatory variables used for scoliosis were: windswept hips, immobility in lying position (PPAS level < 4), having only one lying position, inability to passively straighten legs, and spending >12 h in lying. Explanatory variables used for windswept hips were: scoliosis, immobility in lying position (PPAS level < 4), having only one lying position, inability to passively straighten legs, and spending >12 h in lying. The results were presented as odds ratios (OR), which are ratios between two odds that an event will occur, with 95% confidence intervals (95% CI). SAS Enterprise Guide 7.11 was used for the statistical analyses (SAS Institute Inc., Cary, NC). The first step in the logistic regression process was to calculate estimates of unadjusted OR for each outcome variable and all the explanatory variables. Those explanatory variables, that demonstrated significant unadjusted OR for each of the outcome variable, were used in a backward elimination regression process. For each of the outcome variables, the explanatory variables with the least non-significant p values were eliminated from the next regression model, until only significant values were left in the model. As the variables “passively straighten hips” and “passively straighten knees” were strongly correlated, only ability to “passively straighten knees” were used in the multiple regression models to avoid collinearity problems.

Results

In all, 830 adults with CP participated in the study (469 men and 361 women) at a median age of 23 years (range 16–73 years). The subjects' GMFCS ranged from level I ($n = 159$), II ($n = 185$), III ($n = 130$), IV ($n = 155$) to level V ($n = 201$). The distribution of participants' neurological subtypes was as follows: spastic unilateral ($n = 169$), spastic bilateral ($n = 461$), ataxic ($n = 29$), dyskinetic ($n = 103$), and mixed or unclassified subtypes ($n = 69$). Of the 830 adults in the CPUP database, 228 (27%) lay solely in one position when in bed, 135 (16%) were immobile in lying position, in 216 (26%) passive straightening of the hips (to zero degrees) was not possible and in (449 (54%) passive straightening of the knees was not possible (to zero degrees).

Of the 830 adults, 119 (14%) individuals had a scoliosis, 175 (21%) had windswept hips, and 35 (4%) individuals had both windswept hips and a scoliosis. With respect to passive straightening of hips and knees to zero degrees, passive straightening was not possible in 215 (26%) of hips and in 445 (54%) of knees. GMFCS levels showed a moderate correlation with scoliosis ($r_s = -0.39$, $p < 0.001$) but the correlation with windswept hips was weak ($r_s = -0.27$, $p < 0.001$).

A majority of those with scoliosis were classified at GMFCS level V and spent more than 8 h lying (Table 1). Almost one-third of individuals with scoliosis had solely one lying position, where the most frequent position was side-lying on the right side. In the majority of the individuals with scoliosis the knees could not be straightened passively (due to contracture) and half of the group with scoliosis were unable to change their lying position or passively straighten their hips (due to contracture) (Table 1).

The highest number of individuals with windswept hips was at GMFCS level V and the majority of those with windswept hips spent more than 8 h lying daily (Table 2). Of those with windswept hips, 21% had only one lying position, where the most frequent position was side-lying. In half the individuals with windswept hips, passive straightening of the knees was not

Table 1. Distribution of adults with scoliosis at GMFCS-levels I–V, relative to their time spent in lying, lying position, hip and knee range of motion, and inability to move or change position as measured with PPAS.

	GMFCS level					Total number
	I	II	III	IV	V	
Scoliosis	2	6	12	29	70	119
Time in lying < 8 h	1	3	3	2	1	10
Time in lying 8–12 h	1	3	8	25	53	90
Time in lying > 2 h	0	0	1	2	16	19
One lying position	0	3	5	12	17	37
Supine	0	2	2	1	4	9
Prone	0	0	0	5	5	10
Side-lying (right)	0	0	3	6	3	12
Side-lying (left)	0	1	0	0	5	6
Lack of hip extension	0	1	5	11	33	50
Lack of knee extension	0	4	9	20	32	65
PPAS ability 1–3 (unable to change position)	0	0	1	3	53	57

GMFCS: gross motor function classification system; PPAS: posture and postural ability scale.

Table 2. Distribution of adults with windswept hip distortion at GMFCS-levels I–V, relative to their time spent in lying, lying position, hip and knee joint range of motion, and inability to change position as measured with PPAS.

	GMFCS level					Total number
	I	II	III	IV	V	
Windswept hip distortion	7	34	30	41	63	175
Time in lying < 8 h	1	3	3	2	1	10
Time in lying 8–12 h	1	3	8	25	53	90
Time in lying > 12 h	0	0	1	2	16	19
One lying position	0	2	3	13	17	35
Supine	0	1	2	1	4	8
Prone	0	0	0	5	5	10
Side-lying (right)	0	0	1	6	3	10
Side-lying (left)	0	1	0	0	5	6
Lack of hip extension	0	1	6	11	33	51
Lack of knee extension	0	4	9	22	53	88
PPAS ability 1–3	0	0	1	3	53	57

GMFCS: gross motor function classification system; PPAS: posture and postural ability scale.

Table 3. Unadjusted odds ratios (OR) values of having scoliosis for nine independent factors.

Effect (n)	OR	95 % CI	
Windswept (96)	2.18	1.38	3.45
Supine lying (119)	0.84	0.39	1.83
Prone lying (119)	1.89	0.90	3.98
Side lying (119)	0.94	0.61	1.45
Time lying (119)			
> 12 h vs. < 8 h	7.55	3.25	17.53
8–12 h vs. < 8 h	3.27	1.66	6.44
Lack of hip extension (106)	2.78	1.82	4.25
Lack of knee extension (116)	2.99	1.90	4.70
PPAS lying < 4 (119)	8.39	5.39	13.06

PPAS: posture and postural ability scale.

possible and in 30% passive straightening of the hips was not possible (due to contracture in both cases). Of those with windswept hips, 30% were unable to change position (Table 2). Four of the seven with windswept hips at GMFCS level I, were classified as spastic unilateral.

Of the nine factors tested for association with scoliosis (Table 3), individuals who were immobile in lying position (PPAS < 4), had the highest unadjusted OR (8.4) of having scoliosis, when compared to those who had some movement in lying position. Those who only used one lying position did not have higher odds of scoliosis, compared to those who used alternative

Table 4. Adjusted odds ratios (OR) values of having scoliosis for factors with significant adjusted OR.

Effect (n)	OR	95% CI	
PPAS lying <4 (116)	5.68	3.53	9.14
Lack of knee extension (116)	1.84	1.12	3.00
Time lying (116)			
>12 h vs. <8 h	2.93	1.16	7.39
8–12 h vs. <8 h	2.20	1.08	4.44

PPAS: posture and postural ability scale.

Table 5. Unadjusted odds ratios (OR) of having windswept hips for nine independent factors.

Effect (n)	OR	95% CI	
Scoliosis (160)	2.18	1.38	3.45
Supine (175)	2.18	1.24	3.83
Prone (175)	1.34	0.65	2.76
Side lying (175)	0.83	0.57	1.21
Time lying (170)			
>12 h vs. <8 h	3.30	1.69	6.44
8–12 h vs. <8 h	1.01	0.66	1.56
Lack of hip extension (160)	1.99	1.37	2.90
Lack of knee extension (174)	1.97	1.38	2.81
PPAS lying <4 (175)	3.60	2.36	5.51

PPAS: posture and postural ability scale.

Table 6. Adjusted odds ratios (OR) of having windswept hips for factors with significant adjusted OR.

Effect (n)	OR	95% CI	
PPAS lying <4 (174)	2.90	1.86	4.53
Supine (174)	1.86	1.03	3.34
Lack of knee extension (174)	1.58	1.09	2.29

PPAS: posture and postural ability scale.

lying positions. When adjusting the scoliosis OR values, for all other factors having a significant adjusted OR value (Table 4), those who could not change position independently still had the highest OR (5.7). In addition, in those individuals where passive straightening of the knees was not possible and in those who spent more than 8 h daily in lying, the adjusted OR values for scoliosis were significant.

Of the nine factors tested for association with windswept hips (Table 5), individuals who were immobile in lying position (PPAS <4), had the highest unadjusted OR (3.6), compared to those who had at least some movement in a lying position. Those who lay solely in supine position; also had higher odds of windswept hips compared with those who used other positions. When adjusting the OR values of windswept hips, for all other factors having a significant adjusted OR value (Table 6), those who were unable to change position in lying, passively straighten knees and who lay only in a supine position, had significant OR for windswept hips. GMFCS level, which is a predictor of scoliosis (OR =2.6, 95% CI =2.1–3.1 for each increase in level by one) and windswept hips (OR =1.6, 95% CI =1.4–1.8 for each increase in level by one), was left out of the regression analysis, as the impact of other predictors, on scoliosis and windswept hips, were the focus of this study.

Discussion

The foremost finding of this study indicates that adults with CP who are immobile in a lying position have higher odds of both scoliosis and windswept hips than those who are more mobile. Lying (in one or more positions) for more than 8 h, increased the odds of having scoliosis, while lying solely in a supine position,

gives higher odds of windswept hips. The results from this study are consistent with the findings, in previous work [23,24] in babies and young children with disabilities, that inability to change position in lying results in higher odds of developing both scoliosis and windswept hips. Those who spend longer time in lying, have higher odds of scoliosis.

It has been suggested that side-lying increases the risk and severity of scoliosis [9] due to instability resulting from a narrow base of support and the degree of mobility between body segments. The advice given is to avoid side lying in immobile individuals and use the supine position instead. The results of this study do not support the view that individuals with CP, who only lie on their side have higher odds of scoliosis, than those who only lie supine or use alternative positions. Knee contracture with inability to passively straighten the knees (to zero degrees) was the most common secondary complication of the body in this study. Those whose knees could not be straightened passively have higher odds of scoliosis and windswept hips. In the disabled person with CP lying supine and unable to move or change position, the knees tend to flex and fall to the same side resulting in tissue adaptation, established contracture over time and predisposing to windswept hips. Even the smallest unilateral flexion contracture of a knee, creates an apparent leg length discrepancy, which is standing and walking, leads to an oblique pelvis and eventual scoliosis.

Only a small difference was found between the 21% prevalence of windswept hips in this study and the previously reported prevalence (18%) of windswept hips in children with CP [15]. This study included both ambulatory and non-ambulatory individuals with CP, which is a novelty in studies of windswept hips. Although unexpected, seven individuals at GMFCS level I had windswept hips, according to the range of movement criteria used in this study, which suggests, that a more specific definition of windswept hips is required. Neither Lonstein and Beck [12] nor Persson-Bunke et al. [14] included a rotation of pelvis to the same side as the knees, in their definition of windswept hips. The findings in this study support the conclusion of Hägglund et al. [15] that development of windswept hips is caused initially by knee flexion with fall of the legs to one side in a supine position. Thus, it is not the knee flexion per se, that causes the windswept hips and it is likely the persistent fall of the knees to one side which, in consequence, rotates the pelvis to the same side.

In this study, the prevalence for scoliosis was found to be 14% in adults with CP compared with 11% in the total Swedish population of children with CP [16]. It was unexpected to find that only 30% of the individuals with scoliosis also had windswept hips, compared to 43% in Young et al.'s study [13], resulting in a weaker correlation between scoliosis and windswept hips than anticipated. This may be explained by the CPUP surveillance programme, as it was founded to prevent hip dislocations. The lower number of windswept hips, among individuals with scoliosis, might be partially explained by the reduced number of hip dislocations [25].

Conclusion

The preferred posture, frequently observed in immobile adults with CP, leads to a distortion of their body shape. This posture becomes habitual and is compounded with time in lying and gravity. The results strengthen the need for adequate and appropriate postural support in lying from a young age, in individuals unable to change their posture and position.

Disclosure statement

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