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ORIGINAL ARTICLE

Epidemiology of scoliosis in cerebral palsy: A population-based study at skeletal maturity

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Aim: This study investigated the prevalence of scoliosis in a large, population-based cohort of individuals with cerebral palsy (CP) at skeletal maturity to identify associated risk factors that may inform scoliosis surveillance.

Methods: Young people with CP born between 1990 and 1992 were reviewed through routine orthopaedic review or a transition clinic. Classification of CP was recorded by movement disorder, distribution, gross and fine motor function. Clinical examination was undertaken and those with clinical evidence of scoliosis or risk factors had radiographs of the spine. Scoliosis severity was measured and categorised by Cobb angle.

Results: Two hundred and ninety-two individuals were evaluated (78% of the birth cohort) at a mean age of 21 years, 4 months (range 16–29 years). Scoliosis (Cobb angle $>10^\circ$) was found in 41%, with strong associations to the Gross Motor Function Classification System (GMFCS), Manual Abilities Classification System (MACS) and dystonic/mixed movement disorders. Those at GMFCS V were 23.4 times (95%CI 9.9–55.6) more likely to develop scoliosis than those at GMFCS I. Severe curves (Cobb $>40^\circ$, 13% of the cohort) were found almost exclusively in those functioning at GMFCS IV and V, and were 18.2 times (95%CI 6.9–48.5) more likely to occur in those with dystonia than those with spasticity.

Conclusions: Scoliosis was very common in young people with CP, with prevalence and severity strongly associated with GMFCS and MACS level and dystonic movement disorder. Severe curves were almost exclusively found in non-ambulant children. Clinical screening for scoliosis should occur for all children with CP, with radiographic surveillance focusing on those functioning at GMFCS IV and V.

Key words: cerebral palsy; GMFCS; MACS; scoliosis; surveillance.

What is already known on this topic

- 1 Scoliosis is common in children with cerebral palsy (CP).
- 2 Severe scoliosis is reported to be more common in non-ambulant children.
- 3 There is a lack of clinical guidelines for scoliosis surveillance in CP.

What this paper adds

- 1 This study is population-based with follow-up to skeletal maturity.
- 2 Severe scoliosis is associated with the Gross Motor Function Classification System, Manual Abilities Classification System and movement disorder.
- 3 These findings have implications for scoliosis surveillance in CP.

Cerebral palsy (CP) is the most common cause of physical disability in most developed countries, with a prevalence ranging from 1.5 to greater than 3 per 1000 live births.¹ The widely accepted international definition is

*Cerebral palsy describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, perception, cognition, communication and behaviour, by epilepsy and by secondary musculoskeletal problems.*²

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This definition highlights the musculoskeletal problems which occur secondary to the brain lesion and are known to be progressive as the child grows and develops.^{1–3} Children with CP do not have contractures, hip displacement or scoliosis at birth; these musculoskeletal problems develop over time.^{1–3}

The epidemiology of hip displacement in CP has been well studied, with several population-based studies demonstrating a strong association with the Gross Motor Function Classification

System (GMFCS),^{4–7} and this knowledge has become the key-stone of guidelines for hip surveillance in Australia⁷ and many other countries. We have also previously reported on an inception cohort of children with CP, born in the years 1990–1992, including risk of hip displacement and long-term hip health outcomes associated with hip surveillance.^{5,8,9} Better understanding of the epidemiology of hip displacement in children with CP has informed the implementation of hip surveillance programmes, leading to improved outcomes.^{3,9}

Most studies of scoliosis in children with CP have been based on cohorts referred to outpatient services or have included young people in institutional accommodation.^{10,11} Neither of these methods are able to determine the true prevalence of musculo-skeletal pathologies and associated risk factors. To date, there have been only two population-based studies of scoliosis reported in the literature, based on a large cohort of young people followed by the Cerebral Palsy Follow-up Programme (CPUP) designated a National Quality Register, Sweden.^{12,13} At long-term follow-up, 77% of young people with CP had a clinical assessment of their spine and 13% had a radiographic examination.¹³ The authors reported increasing prevalence and severity of scoliosis with increasing GMFCS level and age.^{12,13} Associations with upper limb function were not studied and associations with CP subtype were not significant.¹³

The aim of this study was to investigate the prevalence of scoliosis in a population of children with CP at skeletal maturity. We aimed to identify the incidence and severity of scoliosis, and to determine associations and risk factors.

Table 1 Classification of the study cohort ($n = 292$) by Gross Motor Function Classification System (GMFCS), Manual Ability Classification System (MACS), movement disorder and topographical distribution

	<i>n</i> (%)
GMFCS	
I	96 (33)
II	46 (16)
III	43 (15)
IV	51 (17)
V	56 (19)
MACS	
I	113 (39)
II	59 (20)
III	47 (16)
IV	33 (11)
V	40 (14)
Movement disorder	
Spastic	156 (53)
Mixed hypertonia	97 (33)
Dystonia	31 (11)
Ataxia	8 (3)
Topographical distribution	
Hemiplegia	81 (28)
Diplegia	85 (29)
Triplegia	11 (4)
Quadriplegia	115 (39)

Methods

An inception cohort of children with CP born in the years 1990–1992 has been described in detail elsewhere,⁸ including long-term follow-up conducted through a transition clinic.¹⁴ In this study, assessment of scoliosis was conducted at the young person's routine orthopaedic review or they were invited to attend a transition clinic appointment. Medical and radiographic history was reviewed and a detailed clinical examination was performed. Classification of CP was recorded by movement disorder and topographical distribution, gross motor function (GMFCS) and fine motor function (Manual Ability Classification System, MACS) according to widely used systems and descriptors.^{1,4,15,16}

Clinical examination of the hips and spine was performed for all children, and radiographs were obtained using standardised techniques.^{3,17} To prevent unnecessary radiation exposure in those without any clinical evidence of scoliosis, repeat radiographic examination at the skeletal maturity review was limited in individuals with hemiplegia to those with Winters, Gage and Hicks type IV gait; a leg length discrepancy >2.5 cm; and/or clinical evidence of asymmetry or scoliosis during a forward bend test.^{3,17} In children with diplegia, a radiograph was obtained for those with identified risk factors of hip displacement, asymmetric involvement, leg length discrepancy >2.5 cm and/or asymmetry noted on a forward bend test.³ All adolescents at GMFCS levels IV and V had a spine radiograph because of the high clinical prevalence of spinal deformity in this group.^{3,13,17} Past history of orthopaedic surgery for both hip displacement and scoliosis were recorded.

This study was approved by Human Research Ethics Committee, The Royal Children's Hospital.

Scoliosis assessment

The forward bend test was used for ambulant patients and those who could stand with minimal support.³ Anteroposterior and lateral radiographs including the entire spine and both hips were obtained. Radiographs were performed in standing position for ambulant children and in sitting or in supine position for non-ambulant children.^{3,17} The Cobb angle¹⁸ from the patient's most

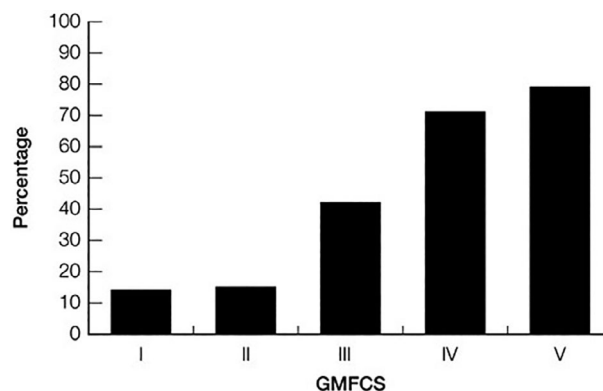


Fig 1 Scoliosis, defined as Cobb angle $>10^\circ$ (■), by the Gross Motor Function Classification System level.

Table 2 Results of logistic regression analysis with scoliosis (Cobb angle >10°) as the outcome

	<i>n</i> (% scoliosis)	Univariate analysis		Multivariate analysis adjusted for GMFCS	
		OR (95% CI)	<i>P</i> value	OR (95% CI)	<i>P</i> value
GMFCS					
I	96 (14)	1	NA	NA	NA
II	46 (15)	1.1 (0.4–3.1)	0.79	NA	NA
III	43 (42)	4.6 (2.0–10.7)	<0.001	NA	NA
IV	51 (71)	15.3 (6.6–35.5)	<0.001	NA	NA
V	56 (79)	23.4 (9.9–55.6)	<0.001	NA	NA
MACS					
I	113 (12)	1	NA	1	NA
II	59 (32)	3.7 (1.6–8.1)	0.001	3.3 (1.4–7.5)	0.004
III	47 (57)	10.4 (4.6–23.5)	<0.001	4.8 (1.6–14.7)	0.01
IV	33 (79)	28.6 (10.4–78.8)	<0.001	15.3 (2.8–84.6)	0.002
V	40 (83)	36.3 (13.3–98.5)	<0.001	26.2 (3.4–200)	0.002
Movement disorder					
Spastic	156 (19)	1	NA	1	NA
Mixed hypertonia	97 (65)	8.1 (4.5–14.5)	<0.001	2.9 (1.4–6.0)	0.01
Dystonia	31 (81)	18.2 (6.9–48.5)	<0.001	7.9 (2.6–23.6)	<0.001
Ataxia	8 (13)	0.6 (0.1–5.3)	0.67	0.7 (0.1–6.4)	0.76

CI, confidence interval; GMFCS, Gross Motor Function Classification System; MACS, Manual Abilities Classification System; NA, not applicable; OR, odds ratio

recent or final radiograph was measured, with the pre-operative radiograph evaluated for the 27 patients who had undergone scoliosis surgery. Measurement was undertaken within the Synapse picture archiving and communication system (Fujifilm Corporation, Tokyo, Japan) in a standardised method by a trained observer.^{3,17,18} Measures were checked by a second observer, a consultant orthopaedic surgeon with extensive experience in the management of neuromuscular scoliosis. The apical vertebra and side of the curve were also noted.

Statistical methods

Descriptive statistics were explored for categorical variables. The severity of scoliosis was classified into three categories: 'no scoliosis' (Cobb angle <10°), 'mild to moderate scoliosis' (Cobb angle ≥10° and <40°) or 'severe scoliosis' (Cobb angle ≥40°). Logistic regression analysis was performed including a univariate analysis with Cobb angle >10° as the outcome, followed by a multivariate analysis adjusted for GMFCS level, for associations between scoliosis, GMFCS, MACS and movement disorder. Odds ratios (ORs), confidence intervals and *P* values were calculated for Cobb angle >10°. Because curves with Cobb angle >40° were only seen at GMFCS IV and V, ORs could not be calculated, and results were expressed as raw numbers and percentages. *P* values of <0.05 were considered statistically significant. Statistical analysis was performed in SPSS software (SPSS v20; IBM, Chicago, IL, USA) and supervised by a biomedical statistician.

Results

Two hundred and ninety-two individuals (170 males and 122 females) were available for review, representing 78% of the birth cohort. Mean age was 21 years, 4 months (range 16–

29 years). Classification by GMFCS, MACS, movement disorder and topographical distribution are summarised in Table 1. With respect to previous descriptions of this cohort, both the topographical distribution and GMFCS level remained stable in the majority of individuals.^{5,8} However, the prevalence of both mixed tone and dystonia was increased according to the criteria described by Sanger *et al.*¹⁵ An exception to GMFCS stability was an 18-year-old girl previously described as having a spastic hemiplegia, GMFCS level II. At age 12, she presented with progressive gait disturbance and was evaluated in the gait laboratory, at which time scoliosis was first noted. Her function had deteriorated to GMFCS level III, and her movement disorder was dystonic. Her scoliosis progressed rapidly, and at the time of surgical correction her Cobb angle was 88° and she was functioning at GMFCS level IV. Following successful scoliosis surgery, her function improved to GMFCS level III.

Scoliosis prevalence

Scoliosis (Cobb angle >10°) was found in 41% of the population with a strong association between GMFCS level, MACS level, movement disorder (CP subtype) and the risk of scoliosis (Fig. 1, Table 2). Compared to GMFCS I, the Odds Ratio, OR for having a curve >10° was 1.1 at GMFCS II, 4.6 at GMFCS III, 15.3 at GMFCS IV and 23.4 at GMFCS V. The relationship between increasing MACS and scoliosis was somewhat similar (Table 2). Compared to spasticity, the movement disorders associated with scoliosis were dystonia (OR 18.2, 95%CI 6.9–48.5) and mixed hypertonia (OR 8.1, 4.5–14.5).

Scoliosis severity

Scoliosis (Cobb angle >10°) was found at all GMFCS levels; however, the majority of curves at GMFCS levels I and II were small and



Fig 2 Standing spine radiograph of an 18-year-old female with spastic diplegia, functioning at the Gross Motor Function Classification System III and Manual Abilities Classification System level II. She has an asymptomatic, C-shaped thoracolumbar scoliosis measuring 30°, with adequate balance of her shoulders over her pelvis, requiring no surgical intervention.

either non-progressive or very slowly progressive (Figs 1,2). In contrast, curves $>40^\circ$ were found only at GMFCS levels IV and V (Figs 3,4), 13% of the whole cohort and 35% of non-ambulant children, with the exception of the patient described previously. In young people functioning at GMFCS IV, 9 of 51 (18%) had a curve $>40^\circ$, and in those at GMFCS V, 27 of 56 (48%) had a curve $>40^\circ$. The majority of young people with curves $>40^\circ$ had severe upper limb involvement (MACS levels III–V) and either dystonia or mixed movement disorder (Table 2). The curve apex was located in the lumbar spine in 57% of young people (22% with apex at L3), with 43% having apex in the thoracic spine.

Twenty-nine young people progressed to surgical correction of their scoliosis by posterior instrumented spinal fusion. Mean age at surgery was 14 years (range 10–23 years) with a mean pre-operative

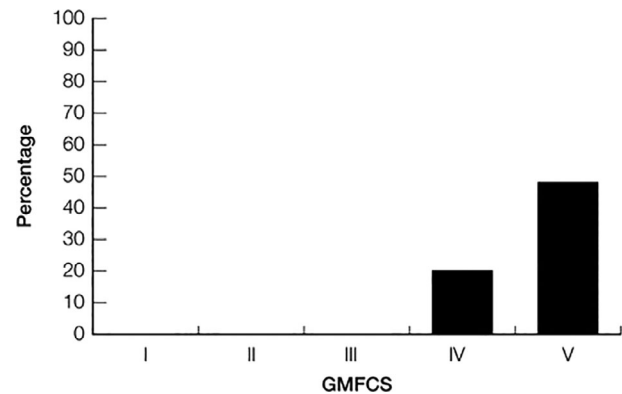


Fig 3 Severe scoliosis, defined as Cobb angle $>40^\circ$ (■), by the Gross Motor Function Classification System level.

Cobb angle of 81° (range 47 – 152°). Some patients with severe scoliosis did not have surgery due to frailty or parent/carer choice.

Discussion

This is the first population-based study of scoliosis in children with CP with follow-up to skeletal maturity and measurement using standardised radiographic measures. As such, it provides insights as to the risk factors associated with the development of mild and severe scoliosis, including gross motor function, upper limb function and movement disorder (Fig. 5).

A scoliosis (Cobb angle $>10^\circ$) was common, affecting 41% of this population of young people with CP. Although mild curves were present at *all* GMFCS levels, the prevalence and severity of scoliosis showed a strong relationship to GMFCS, MACS and movement disorder (Fig. 5). The majority of curves at GMFCS levels I–III were small and mostly non-progressive, with minimal or no symptoms (Fig. 2), and may have been related to mild leg length inequality or postural factors.^{3,19} We suggest that small curves in ambulant children require clinical monitoring, with radiographic examination only when indicated on clinical grounds. Conversely, curves $>40^\circ$ occurred almost exclusively at GMFCS levels IV and V. Compared to those with function at GMFCS I, those at GMFCS V were 23 times more likely to develop severe scoliosis. Importantly, the prevalence of severe scoliosis was twice as high in children at GMFCS V compared to GMFCS IV (Fig. 3). When adjusted for GMFCS, those with upper limb function at MACS level V were 26 times more likely to develop scoliosis.

Our findings support those of colleagues in Sweden, who noted that the incidence of scoliotic curves $>40^\circ$ increased with increasing GMFCS and age.^{12,13} Our study had a higher utilisation of radiographic assessment than the Swedish studies, which permitted more rigorous assessment of the prevalence and magnitude of scoliosis in young people who are ambulant.^{3,12,13,17} Parents of ambulant young people were frequently concerned about the impact of minor limb length inequality and postural asymmetry on spinal alignment. The low prevalence of scoliosis in this group found in our study and the CPUP studies are reassuring in this regard.^{12,13}

As noted in previous studies, curves may start in early childhood, can progress rapidly and have a negative impact on sitting

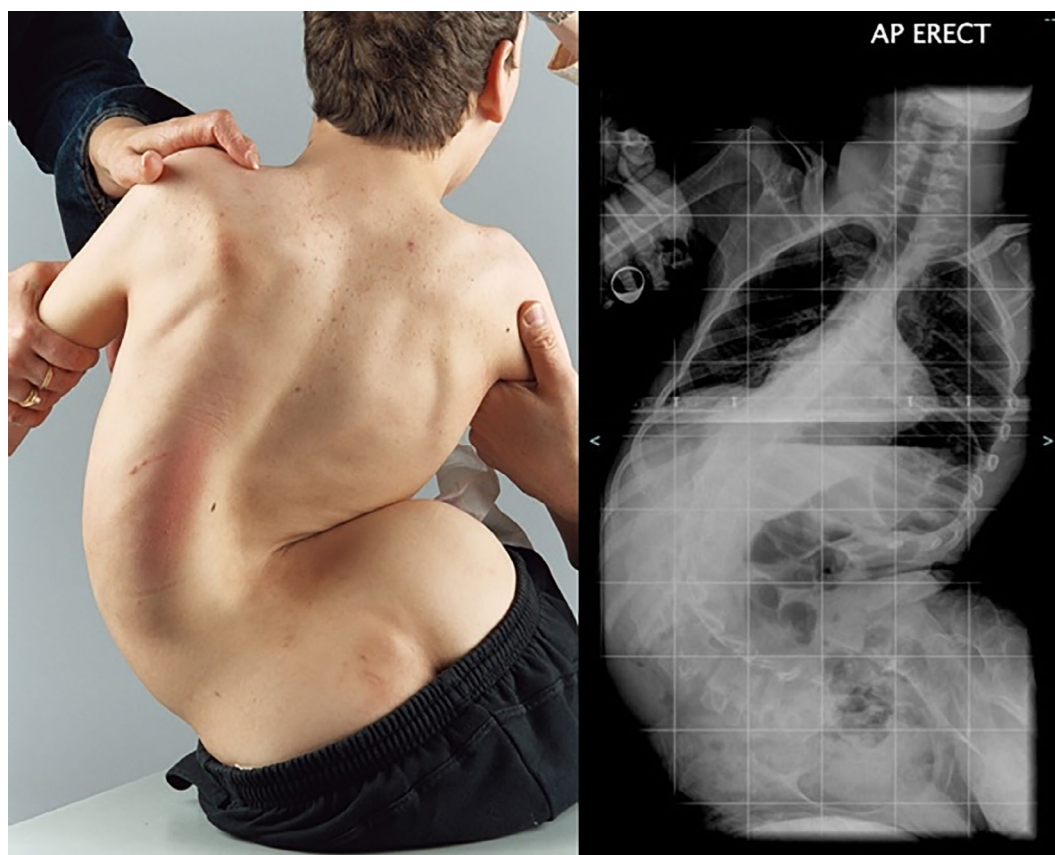


Fig 4 Clinical photograph and spine radiograph of a 16-year-old male functioning at the Gross Motor Function Classification System and Manual Abilities Classification System level V, with a mixed spastic–dystonic movement disorder. There is a long C-shaped thoracolumbar scoliosis, measuring 110°. There is severe pelvic obliquity and loss of sitting ability. Note the head and trunk support required to acquire a radiograph and clinical photography in a sitting position.

ability, comfort and care by the second decade.^{19–21} In non-ambulatory children, age and curve severity have been identified as predictors of curve progression. In particular, a curve $>40^\circ$ by age 12 was more likely to progress. This study is the first to identify impaired upper limb function and movement disorder type as risk factors for severe scoliosis. The pathogenesis of scoliosis in CP is complex and probably multifactorial (Fig. 5). Higher GMFCS levels (IV and IV) are associated with core weakness, impaired sitting balance and head control, generalised hypertonia and lack of mobility. Higher MACS levels are associated with impaired use of the upper limbs for sitting balance and to adjust sitting position. These associations may point to possible preventive strategies such as supportive seating, bracing and management of hypertonia.^{19,22}

In non-ambulant individuals, scoliosis may continue to progress during adolescence and adult life and may lead to pain, loss of sitting, impaired quality of life, respiratory compromise and death from respiratory failure.^{10,11,21,23} The only intervention which has been shown to be successful for severe curves in CP is orthopaedic surgery.¹⁷ Surgical management may be considered for children who cannot maintain sitting posture due to pain or deformity, have fixed pelvic obliquity with a hip at risk or have risk of developing decubitus ulceration or respiratory compromise.^{3,17,19,20,23} While surgical correction of scoliosis in CP has

been reported to improve health-related quality of life,²³ children with CP have a higher rate of adverse events following surgery, including blood loss, infection, pulmonary complications and mortality, particularly when a curve exceeds 60° .^{17,19,20,23,24} Effective multidisciplinary care, discussion and shared decision-making with parent and caregivers is important in exploring when, or if, to proceed with scoliosis surgery.

Hip surveillance is effective in improving hip health outcomes at skeletal maturity through early identification of progressive hip displacement and facilitating timely orthopaedic referral.^{3,9} While not all children require surgical management, early referral creates opportunity and time for multidisciplinary assessment and care, information sharing and shared decision-making between parents and caregivers and careful perioperative preparation for children who progress to hip surgery.^{3,9} It is critical that a similar process of surveillance, early identification and timely orthopaedic referral also occurs with regard to scoliosis.¹³ While evidence for the effectiveness of non-operative management of scoliosis is variable, early identification may allow opportunity to trial these strategies where parents prefer, or for children for whom surgical risks may be high.^{3,13,22} Moreover, early identification and referral allows time for discussion, decision-making and planning to occur before a severe scoliosis develops and the risk of surgical complications increases.^{12,13,24}

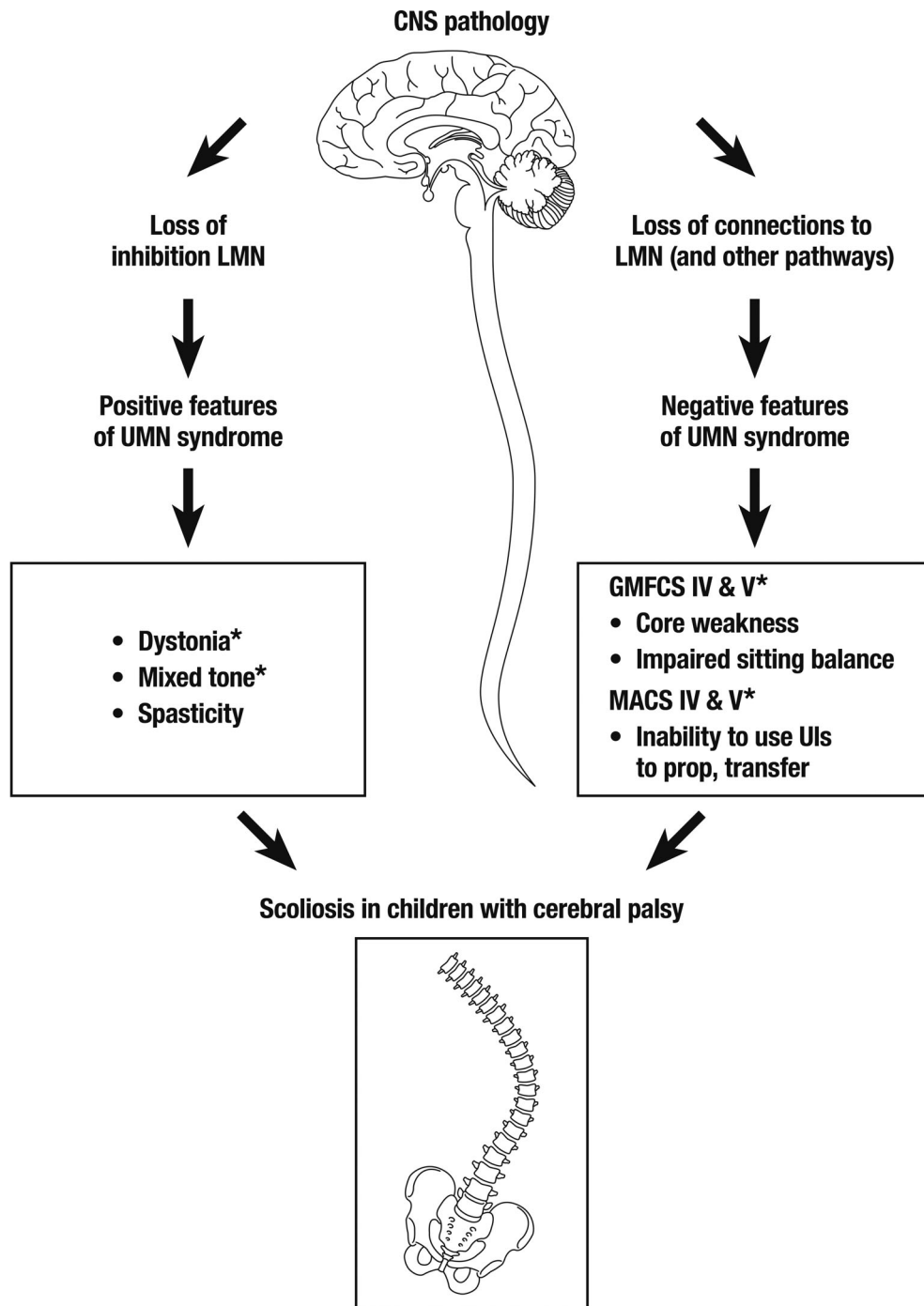


Fig 5 Pathogenesis of scoliosis in CP. The injury to the UMNs in CP results in both ‘positive’ and ‘negative’ clinical features. The predominant negative features result from loss of corticospinal tract connections to LMNs causing paresis or partial paralysis of skeletal muscle. By contrast, hypertonia is hypothesised to be caused by the loss of inhibitory descending input to the LMNs, which impedes overactivity in the stretch reflex in the peripheral neuromuscular system. This loss results in hypertonia and hyper-reflexia. The effects of the brain lesion in children with CP may extend to all parts of the musculoskeletal system, with scoliosis being one of the common musculoskeletal deformities in children with more severe motor impairment (GMFCS IV and V). Both positive (too much tone) and negative (too little selective motor control and strength) features of the UMN syndrome are associated with the development of severe scoliosis (Cobb angle $>40^\circ$). *Factors with a statistically significant association with development of severe scoliosis, Cobb angle $>40^\circ$ (CNS, central nervous system; CP, cerebral palsy; GMFCS, Gross Motor Function Classification System; LMN, lower motor neuron; MACS, Manual Abilities Classification System; UMN, upper motor neuron).

Our study was limited by follow-up which was shorter than ideal in some young people and with very few clinical and radiographic assessments for some of those at high risk. Our data collection also limited formal exploration of age at onset of scoliosis, or risk factors associated with more rapid progression. The strengths of the study were systematic clinical and radiographic follow-up to skeletal maturity or beyond in a large percentage of the population, along with review and classification of GMFCS, MACS levels and movement disorder classification by a small number of experienced observers.

Conclusions

Mild scoliosis is common in individuals with CP at all GMFCS levels, but curves $>40^\circ$ develop almost exclusively in those who are non-ambulatory (GMFCS IV and V), have severe fine motor impairment (MACS IV and V) and dystonia or mixed movement disorder. We strongly recommend that although regular clinical screening for scoliosis should occur for all children with CP, radiographic surveillance must focus on those functioning at GMFCS IV and V. It is imperative that a synthesis of these findings and those of previous studies is now undertaken to inform the development of detailed clinical guidelines for the surveillance of scoliosis in young people with CP.

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