Demographic and modifiable factors associated with knee contracture in children with cerebral palsy

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This article is commented on by Van Campenhout and Bar-On on pages 335–336 of this issue.

PUBLICATION DATA
Accepted for publication 29th November 2017.
Published online 10th January 2018.

ABBREVIATIONS

CPUP	Cerebral Palsy Surveillance Programme
ROM	Range of motion

AIM To identify the prevalence of knee contracture and its association with gross motor function, age, sex, spasticity, and muscle length in children with cerebral palsy (CP).
METHOD Cross-sectional data for passive knee extension were analysed in 3 045 children with CP (1 756 males, 1 289 females; mean age 8y 1mo [SD 3.84]). CP was classified using the Gross Motor Function Classification System (GMFCS) levels I (n=1 330), II (n=508), III (n=280), IV (n=449), and V (n=478). Pearson's χ^2 test and multiple binary logistic regression were applied to analyse the relationships between knee contracture and GMFCS level, sex, age, spasticity, hamstring length, and gastrocnemius length.
RESULTS Knee contracture greater than or equal to 5 degrees occurred in 685 children (22%). The prevalence of knee contracture was higher in older children and in those with higher GMFCS levels. Odds ratios (ORs) for knee contracture were significantly higher for children at GMFCS level V (OR=13.17), with short hamstring muscles (OR=9.86), and in the oldest age group, 13 years to 15 years (OR=6.80).
INTERPRETATION Knee contracture is associated with higher GMFCS level, older age, and shorter muscle length; spasticity has a small effect. Maintaining muscle length, especially of the hamstrings, is important for reducing the risk of knee contracture.

Despite preventive treatment, children with cerebral palsy (CP) often develop muscle contracture that limits their range of motion (ROM). Contracture often results from the chronic activation of a muscle, sometimes in combination with weakness of the antagonists, which occurs in people with CP.[1,2] Low joint mobility also affects adjacent joints and can cause postural asymmetry.[3] The mechanisms underlying contracture in CP are complex and are not fully understood.[4]

Knee flexion contracture frequently occurs in children with CP. A number of different factors can contribute to the development of knee contracture, including older age, spasticity, muscle imbalance, immobility, and prolonged sitting posture.[5,6] Limited knee extension affects both ambulant and non-ambulant children. For non-ambulant children, knee contracture may affect the ability to transfer and to assume an aligned standing or lying posture. For ambulant children, knee contracture can cause an asymmetric posture, impaired gait pattern, increased energy consumption, and difficulty wearing orthoses.[7]

Limited knee extension can lead to a gait pattern that imposes a greater pressure on the knee extensor apparatus and the patellofemoral joint, which can cause knee pain.[8] The proportion of children with pain increases with age and pain is most common in the lower extremities.[9] One in four adults with CP experiences knee pain,[10] which is associated with decreased joint mobility.

The Cerebral Palsy Surveillance Programme (CPUP) is a national quality registry in Sweden.[11] More than 95 per cent of all children with CP nationwide are included in this programme. In the CPUP, children are followed regularly by their local rehabilitation team and are examined every 6months, once a year, or every other year, depending on their age and level in the Gross Motor Function Classification System (GMFCS).[12]

To our knowledge, no studies have investigated multiple factors associated with knee contracture in a large population of children with CP. A deeper understanding of the factors that contribute to the development of knee contracture might help in the creation of treatment strategies.[1] The purpose of this study was to determine the prevalence of knee contracture and its association with GMFCS level, age, sex, spasticity, and muscle length in children with CP.

METHOD

Participants

This was a cross-sectional study based on data from the Swedish CP registry and the CPUP. The study participants were all 3 045 children with CP aged 1 year to 15 years, born between 2000 and 2014 who were reported to the registry in 2014 and 2015. The data for the most recent visit were used. The children represent all 21 counties in Sweden with a total population of 10 million inhabitants. The Research Ethics Committee at Lund University approved the study (LU-443-99), and permission to extract data was obtained from the registry holder.

Classifications and measurements

The diagnosis of CP was verified at the age of four years by a neuro-paediatrician, according to the inclusion and exclusion criteria of the Surveillance of Cerebral Palsy in Europe network.[13] Children younger than 4 years with presumed, although not yet confirmed, CP are included in the CPUP, while those who at a later stage do not fulfil the criteria for CP are excluded. In the present study, 521 children were younger than 4 years with presumed but not yet confirmed CP. Gross motor function was classified by the child's physiotherapist into levels I to V according to the expanded and revised version of the GMFCS.[12]

Passive knee extension was assessed by goniometric measurement in a standardized supine position according to the CPUP manual (http://www.cpup.se). In the CPUP, ROM is graded into three levels according to a traffic light system with separate cut-offs for GMFCS levels I to III compared with GMFCS levels IV to V. Green indicates a good passive ROM. Yellow indicates a reduced ROM and the need for increased observation and actions to improve ROM. Red indicates the development of a contracture that requires intervention. In this study, we used the same classification values for all GMFCS levels. We classified knee extension as a mild contracture if the extension was minus 5 degrees to minus 14 degrees and as a severe contracture if the extension was minus 15 degrees or less.

The hamstring length was classified according to the popliteal angle with the hip flexed to 90 degrees with the child in the supine position. We classified a popliteal angle of 120 to 139 degrees as indicating a mildly reduced hamstring length and below 120 degrees as a severely reduced hamstring length. The length of the gastrocnemius was classified according to the angle of the ankle joint in dorsiflexion with the hip and knee extended. Angles between 9 degrees and 0 degrees of dorsiflexion were considered to indicate mildly reduced muscle length and below 0 degrees indicated severely reduced muscle length.

Muscle tone of the knee and plantar flexors was measured according to the Modified Ashworth Scale.[14] In the statistical analyses, the Ashworth levels were divided into three groups: 0, normal muscle tone; 1 to 1+, slight increase in muscle tone; and 2 to 4, large increase in muscle tone. For all children in this study, information from only one leg, that with the lowest ROM, was used in the analyses. If the ROM was equal on both sides, we used the leg with the highest Ashworth level. For children with the same measurements in both legs, one leg was randomly assigned to be included in the analyses.

Passive knee extension was analysed according to GMFCS level, age, sex, spasticity of the knee and plantar flexors, and length of the hamstring and gastrocnemius muscles.

What this paper adds

- Knee contracture occurs in children with cerebral palsy at all Gross Motor Function Classification System (GMFCS) levels.
- Knee contracture in children is associated with short hamstring muscles, higher GMFCS level, and older age.
- Short hamstring muscles present a greater risk for knee contracture than spasticity.

Statistical analyses

In the statistical analyses, age was classified into five groups (1–3y, 4–6y, 7–9y, 10–12y, and 13–15y). The Ashworth levels were divided into three groups (0, 1 to 1+, and 2 to 4). Knee extension and length of the hamstrings (popliteal angle) and gastrocnemius (dorsiflexion) were also divided into three groups: normal, mild contracture, and severe contracture. Categorical variables are described by frequencies (n) and percentage (%).

Pearson's χ^2 test was used to analyse categorical data, while a χ^2 test for trend (linear-by-linear association test) was used to analyse trends in knee contracture according to GMFCS level and age. Since the outcome variable knee extension was originally measured as the angle of extension, it is essentially a circular data variable, that is, given as the angle of a circle.[15] To fully take this characteristic into account in the statistical analyses, the Watson–Williams test for homogeneity of means was used to identify differences in the mean direction of knee extension between the different levels of categorical variables. For these analyses, knee extension is described by mean direction with accompanying circular standard deviation.

Multiple binary logistic regression was used to analyse the simultaneous associations between the outcome knee contracture (yes/no) and the explanatory variables sex, age, GMFCS level, hamstring and gastrocnemius muscle lengths, and spasticity of the knee and plantar flexors. For the regression analysis, a child was classified as having a knee contracture if the knee extension was below 5 degrees or less (i.e. mild or severe contracture) and as not having a knee contracture if the knee extension was below 4 degrees or more. The results of the regression analyses are presented as odds ratios (ORs) with accompanying 95 per cent confidence intervals (CIs). Nagelkerke R^2 is used as a measure of goodness-of-fit. SPSS v23/24 (IBM Corp., Armonk, NY, USA) and R 3.3.2 (R Foundation for Statistical Computing, Vienna, Austria) with the package 'circular' version 0.4-7 were used for the statistical analyses, with p values less than 0.05 considered significant.

RESULTS

Data were obtained from a total of 3 045 children; 1 756 (58%) males and 1 289 (42%) females. Their mean age was 8 years 1 month (SD 3.84). The distributions according to age, sex, and GMFCS levels are presented in Table I.

Table I: Distribution of age, sex, and GMFCS level

Age (y)	*N* of children	Sex *n* (%)		GMFCS level *n* (%)				
		Male	Female	I	II	III	IV	V
1–3	521	288 (55)	233 (45)	212 (41)	90 (17)	61 (12)	79 (15)	79 (15)
4–6	737	440 (60)	297 (40)	358 (49)	98 (13)	76 (10)	100 (14)	105 (14)
7–9	683	401 (59)	282 (41)	282 (41)	124 (18)	60 (9)	106 (15)	111 (16)
10–12	665	390 (59)	275 (41)	301 (45)	114 (17)	46 (7)	91 (14)	113 (17)
13–15	439	237 (54)	202 (46)	177 (40)	82 (19)	37 (8)	73 (17)	70 (16)
Total	3 045	1 756 (58)	1 289 (42)	1 330 (44)	508 (17)	280 (9)	449 (15)	478 (16)

GMFCS, Gross Motor Function Classification System.

Knee contracture was reported in 685 (22%) of the 3 045 children; 369 (12%) had a mild contracture (−5° to −14°) and 316 (10%) had a severe contracture (−15° or less). Data for passive knee extension were missing for 67 children, 43 of whom were classified as being in GMFCS I. Slightly more males (n=419, 24%) than females (n=266, 21%) had a knee contracture. The percentages of children with a severe contracture were 12 per cent in GMFCS level III, 25 per cent in level IV, and 35 per cent in level V. A severe contracture was exhibited by 3 per cent of children in the 4- to 6-year-old group and 25 per cent of children in the 13- to 15-year-old group (Table II).

Significant differences were observed in knee extension related to sex (p=0.029), age (p<0.001), GMFCS level (p<0.001), length of the hamstring and gastrocnemius muscles (p<0.001), and spasticity of the knee and plantar flexors (p<0.001) (Table III). Knee extension was slightly lower in males than in females; the mean directions (circular SDs) were −2.4 degrees (0.2°) and −1.7 degrees (0.2°) respectively. Knee extension decreased with increasing age from 0.9 degrees (0.1°) for the 1-year to 3-year-old group and to −7.1 degrees (0.2°) for the 13-year to 15-year-old group. Knee extension also decreased with increasing GMFCS level from 1.5 degrees (0.1°) for children at GMFCS level I to −10.6 degrees (0.3°) for those at GMFCS level V. The largest difference in knee extension was related to the length of the hamstring muscle; the mean direction of knee extension (circular SD) was 1.0 degree (0.1°) for children with a normal muscle length compared with −19.5 degrees (0.3°) for those with a severe knee contracture (Table III).

In the multiple binary logistic regression model, age, GMFCS level, hamstring and gastrocnemius muscle lengths, and spasticity of the knee flexors were significantly

Table II: Distribution of knee extension in relation to sex, age, and GMFCS level

Variable	Level	Knee extension: Normal ROM ≥−4° *n* (%)	Mild contracture −5° to −14° *n* (%)	Severe contracture ≤−15° *n* (%)
Sex	Male	1 300 (75)	220 (13)	199 (12)
	Female	993 (79)	149 (12)	117 (9)
Age (y)	1–3	461 (93)	30 (6)	5 (1)
	4–6	611 (85)	85 (12)	25 (3)
	7–9	576 (77)	90 (13)	66 (10)
	10–12	443 (67)	103 (16)	112 (17)
	13–15	262 (61)	61 (14)	108 (25)
GMFCS level	I	1 214 (94)	73 (6)	0 (0)
	II	447 (89)	49 (10)	4 (1)
	III	199 (72)	43 (16)	33 (12)
	IV	236 (53)	96 (22)	112 (25)
	V	197 (42)	108 (23)	167 (35)

GMFCS, Gross Motor Function Classification System; ROM, range of motion.

Table III: Knee extension in relation to sex, age, GMFCS level, hamstring and gastrocnemius muscle lengths, and spasticity of the knee and plantar flexors

Variable	Level	*n* (%)	Knee extension (°), mean direction (CSD)	*p*-value[a]
Sex	Male	1 719 (58)	−2.43 (0.17)	0.029
	Female	1 259 (42)	−1.65 (0.16)	
Age (y)	1–3	496 (17)	0.90 (0.08)	<0.001
	4–6	721 (24)	0.28 (0.11)	
	7–9	672 (23)	−1.68 (0.14)	
	10–12	658 (22)	−4.26 (0.20)	
	13–15	431 (14)	−7.08 (0.24)	
GMFCS level	I	1 287 (43)	1.50 (0.07)	<0.001
	II	500 (17)	0.96 (0.08)	
	III	275 (9)	−2.44 (0.14)	
	IV	444 (15)	−7.13 (0.20)	
	V	472 (16)	−10.58 (0.25)	
Muscle length				
Hamstring muscle	Normal	1 825 (62)	0.97 (0.08)	<0.001
	Mildly shortened	924 (31)	−4.75 (0.17)	
	Severely shortened	189 (6)	−19.52 (0.30)	
Gastrocnemius muscle	Normal	1 744 (59)	−0.77 (0.15)	<0.001
	Mildly shortened	903 (31)	−2.48 (0.14)	
	Severely shortened	287 (10)	−5.61 (0.17)	
Spasticity				
Knee flexors	0	1 751 (69)	0.41 (0.11)	<0.001
	1–1+	548 (22)	−4.11 (0.18)	
	2–4	238 (9)	−10.70 (0.24)	
Plantar flexors	0	1 279 (53)	−0.32 (0.14)	<0.001
	1–1+	674 (28)	−2.04 (0.17)	
	2–4	480 (20)	−5.08 (0.19)	

[a]*p*-values for the difference in mean direction between group levels using the Watson–Williams test for homogeneity of means. CSD, circular standard deviation; GMFCS, Gross Motor Function Classification System.

Table IV: Results of multiple binary logistic regression for the association between knee contracture (yes/no) and sex, age, GMFCS level, hamstring and gastrocnemius muscle lengths, and spasticity of the knee and plantar flexors

Variable	Level	OR	95% CI	*p*-value
Sex	Male	Ref.		
	Female	0.95	(0.71–1.27)	0.714
Age (y)	1–3	Ref.		
	4–6	1.88	(1.02–3.49)	0.044
	7–9	3.27	(1.80–5.94)	<0.001
	10–12	5.18	(2.84–9.45)	<0.001
	13–15	6.80	(3.62–12.77)	<0.001
GMFCS level	I	Ref.		
	II	0.95	(0.58–1.57)	0.849
	III	3.52	(2.14–5.79)	<0.001
	IV	7.91	(5.03–12.46)	<0.001
	V	13.17	(8.42–20.60)	<0.001
Muscle length				
Hamstring muscle	Normal length	Ref.		
	Mildly shortened	3.05	(2.24–4.17)	<0.001
	Severely shortened	9.86	(5.49–17.69)	<0.001
Gastrocnemius muscle	Normal length	Ref.		
	Mildly shortened	1.70	(1.24–2.32)	0.001
	Severely shortened	3.62	(2.31–5.66)	<0.001
Spasticity				
Knee flexors	0	Ref.		
	1–1+	1.49	(1.03–2.15)	0.035
	2–4	2.10	(1.27–3.47)	0.004
Plantar flexors	0	Ref.		
	1–1+	1.00	(0.70–1.42)	0.984
	2–4	0.80	(0.51–1.24)	0.317

Results based on n=2 168 out of n=3 045 individuals with complete cases (complete data set and no missing variables included in the regression analysis), of which 407 (19%) had knee contracture. Nagelkerke R^2=0.499. CI, confidence intervals; GMFCS, Gross Motor Function Classification System; OR, odds ratios; Ref., reference.

associated with an increased risk of knee contracture (Table IV). The goodness-of-fit was quite high, with Nagelkerke R^2 equalling 0.499. However, neither sex nor spasticity of the plantar flexors was significantly associated with knee contracture. Mildly reduced (OR 3.05) and severely reduced (OR 9.86) hamstring length and reduced gastrocnemius length (OR 1.7–3.62) were associated with an increased risk of knee contracture. A higher degree of spasticity in the hamstrings (OR 1.49–2.10) was also associated with a slightly increased risk of knee contracture. Both reduced hamstring length and reduced gastrocnemius length were more strongly associated with knee contracture than increased spasticity in the knee or plantar flexors.

DISCUSSION

Despite the high prevalence of knee contracture in CP, few studies have investigated this issue in children with CP. We found that almost one out of four children of a total population of children with CP aged up to 15 years, which included all GMFCS levels, had a knee contracture of 5 degrees or more. Children at all GMFCS levels exhibited knee contracture, although this was more common in older children and in those with a lower level of motor function. A strong association was observed between reduced hamstring length and knee contracture. Reduced hamstring and gastrocnemius lengths conferred a higher risk for knee contracture than increased spasticity of the knee flexors, but no significant association was found between knee contracture and spasticity in the plantar flexors.

Our finding that 2 per cent of the 13-year to 15-year-olds had knee contractures compared with 10 per cent of the 4-year to 6-year-olds is consistent with previous studies that have reported an increase in the frequency of knee contractures with age.[5,6] It is also the most frequent contracture in adults with CP.[3] In our study, the frequency of knee contracture increased with age at all GMFCS levels, but was more frequent in children with a lower level of motor function. For example, none of the children at GMFCS level I had a severe contracture compared with 35 per cent of those at level V. Even in children at GMFCS level V who lack the ability to walk, knee contracture is highly associated with postural asymmetries, which may cause progressive deformity such as scoliosis, windswept hip deformity, or hip dislocation when the legs are tilted to the side in the supine position. Limited knee extension also obstructs the ability to find optimal lying and standing positions for these children.[3]

Knee contracture was strongly associated with reduced muscle length, particularly of the hamstrings, but also to the gastrocnemius muscles to some extent. In a study by Nordmark et al.[6] hamstring length measured as the popliteal angle started to reduce on average 4 years before the development of knee contractures in children with CP. Both the hamstring and gastrocnemius are two-joint muscles that pass the knee joint and thus affect its ROM. It is common for nonoperative interventions to maintain the length of the gastrocnemius.[16,17] Short hamstring muscles are a challenge and often require operative interventions such as hamstrings release[18] or extension osteotomy. This study shows that children with a hamstring length of less than 120 degrees have an almost 10-fold increase in the risk of knee contracture.

Molenaers et al.[19] described the importance of treating spasticity to prevent joint deformity. However, our data suggest that knee contracture is more strongly associated with reduced muscle length than with spasticity, which may also contribute to reduced muscle length. It is sometimes difficult to distinguish limb stiffness resulting from muscle shortening from that caused by spasticity. Because muscle stiffness increases with age, this difficulty in differentiating shortening from spasticity may occur more frequently in adolescents than in younger children.[20]

This study had several limitations. The Ashworth Scale is a widely used clinical measure of spasticity, but its validity and precision remain open to question.[14] In this study, only 1 of 2 844 children had a reported grade of 4 (rigid) on the Ashworth Scale for the knee flexors, and only 55 (2%) had a reported grade of 3, although more than 10 per cent of the children had a severe knee contracture. Several children with contracture and spasticity received treatment to reduce spasticity, such as the use of an intrathecal baclofen pump and botulinum neurotoxin injections, which may explain the low numbers reported as grade 3 or 4. In the present study, the low reliability of the Modified Ashworth Scale may have underestimated the effects of spasticity on knee contracture. The reliability is slightly higher for the original Ashworth Scale than for the Modified Ashworth Scale and, in this study, we grouped the 1 and 1+ levels according to the original scale.

Another limitation of this study was the cross-sectional design, which involved the collection of information from the last examination for each child and no longitudinal data to follow the development of contracture over time. Our study does not reflect the natural development of knee contracture because many of the children received treatment to prevent its development. Since the CPUP programme started, the number of children with severe contracture has been reduced and the children receive earlier treatment.[21] However, with no treatment, the relationship between reduced muscle length and development of knee contracture would probably have been even more pronounced. The cut-off value of greater than or equal to 5 degrees (mild contracture) affected the high frequency of knee contracture in the present study. A cut-off value of greater than or equal to 15 degrees (severe contracture) would have reduced the percentage by more than half, from 23 per cent to 11 per cent. The measurement of the popliteal angle was used as a surrogate value for hamstring length. Unless adjusted for, the popliteal angle measured in supine with the opposite leg straight may be affected by a limited hip flexion below 90 degrees or a hip flexion contracture pulling the pelvis forward. However only 218 of 3 045 children (7%) were reported to have a hip flexion contracture exceeding 5 degrees on either side.

One strength of this study is that it included a total population of children with CP at all GMFCS levels aged from 1 year to 15 years. Given the prevalence and distributions of sex and GMFCS levels, the study population is likely to be representative of children with CP in other countries with similar development.

The association between knee contracture and reduced hamstring and gastrocnemius muscle lengths is important for the treatment of children with CP. Chan and Miller[18] described the importance of early interventions and noted that children should not be allowed to develop severe contracture that causes them to lose function and ambulatory potential. Maintaining hamstring and gastrocnemius muscle lengths can reduce the risk of developing severe knee contracture and may prevent contracture of the ankle and hip.

In conclusion, knee contracture is frequent in children with CP at all GMFCS levels and is associated with higher GMFCS level, older age, and reduced length of the hamstring and gastrocnemius muscles.

ACKNOWLEDGEMENTS

The authors thank the children and their families who participate in the CPUP.

The authors have stated that they have no interests that might be perceived as posing a conflict or bias.

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