INTRODUCTION

Cerebral palsy (CP) is one of the most common musculoskeletal disabilities globally, with prevalence estimates at 2-3 per 1000 live births.1-3 Spastic CP is the most common subtype, accounting for approximately 75 per cent.1,4 CP can also be classified according to the individual’s gross motor function performance, and the Gross Motor Function Classification System (GMFCS) is used for this purpose.5,6 Comorbidities and secondary conditions affect, to varying degrees, communication, perception, sensation, behaviour, cognition, health and quality of life. These conditions occur frequently and may develop or deteriorate over time.7,8

Bone and joint complications and reduced mobility are associated with pain in children with cerebral palsy

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Abstract
Aim: To investigate the relationships between pain in the lower extremities and back, and spasticity, bone/joint complications and mobility.

Methods: Retrospective population-based registry study. Participants (N = 3256) with cerebral palsy (CP), 2.5-16 years of age, participating in the Swedish Cerebral Palsy Follow-up Program were included. Spasticity was measured using scissoring and the Modified Ashworth Scale. Bone/joint complications consisted of hip displacement, range of motion, windswept posture and scoliosis. Mobility was measured using the Functional Mobility Scale (5-, 50- and 500-metres), wheelchair use (outdoors) and the ability to stand/get up from sitting/use stairs, respectively. Pain was measured as presence of pain in hips, knees, feet and back. Data were analysed using structural equation modelling.

Results: Bone/joint complications had the strongest direct pathway with pain in the lower extremities (standardised regression coefficient = 0.48), followed by reduced mobility (standardised regression coefficient = -0.24). The pathways between spasticity and pain, and age and pain were not significant. The R² of the model was 0.15.

Conclusion: Bone/joint complications and reduced mobility were associated with pain in the lower extremities when controlling for sex. Considering the R² of the model, other factors not included in the model are also associated with pain in the lower extremities in children with CP.

KEYWORDS
Cerebral palsy, pain, spasticity, deformity, mobility, gross motor function
Pain is a frequently reported secondary condition in this population occurring in both sexes, at multiple body sites, and across different GMFCS levels, subtypes and ages. Pain, in this context, tends to be long-lasting, if not chronic,12-15 and can affect important aspects of life such as psychosocial health, participation in recreational activities, educational endeavours and, at older ages, the ability to work.13 In a European multisite study, close to 75 per cent of adolescents with CP self-reported having experienced pain in the previous week.14 In a Swedish population-based study of 2777 children and adolescents with CP, one-third of the population reported pain.15 Pain occurred more frequently in the back and the lower extremities, was more often reported in girls, and increased with age, which has been reported consistently in the literature.14,16,17 Per definition, CP affects motor function and musculoskeletal complications such as hip dislocations, scoliosis, contractures (muscle shortening), spasticity (increased muscle tone), windswept posture and joint complications, all of which can be painful.18 Compromised positioning results in muscle fatigue and may cause pain due to pressure or muscle overuse. Additionally, muscle weakness with or without spasticity and contractures affects mobility. In individuals with CP who are ambulatory, this can be associated with pain primarily related to overuse of the hip, knee and foot extensor muscles.9,15

To prevent musculoskeletal pain from manifesting, a better understanding of the relationships among different factors that cause pain is needed. In this study, we developed models to simultaneously test how pertinent factors are related to pain in the lower body of children and adolescents with CP. More specifically, we investigated the relationships between the presence of pain in the lower extremities (feet, knees and hips) and back, spasticity, bone/joint complications and reduced mobility in relation to age. Higher levels or degrees of spasticity, bone/joint complications, reduced mobility and older age were hypothesised to be associated with an increase in the presence of pain.

2 METHODS

2.1 Procedure and participants

This was a retrospective cohort registry study using data from the Swedish follow-up programme and national registry Cerebral Palsy Follow-up Program (CPUP). All habilitation units in Sweden where children with CP generally receive care participate in CPUP. Children suspected of having CP are eligible to participate, resulting in a population-based database that includes approximately 95 per cent of children with (or suspected) CP born after 2000.9 Data from the most recent CPUP assessments from 2009 to 2017 were included. Participants were included if they were between the ages of 2 years and 6 months and 16 years and 0 months. Exclusion criteria were having undergone selective dorsal rhizotomy, having had a baclofen pump inserted, or not having answered the pain items. The recommended CPUP assessment schedules are based on age and GMFCS level.19 Children at GMFCS level I are examined by physical and occupational therapists annually up to 6 years of age, and then every second year. Those at GMFCS levels II-V are examined twice a year up to 6 years of age, then once a year. Children at GMFCS levels II-V undergo radiographic hip examinations to assess hip migration once a year. However, children at GMFCS level I only undergo radiographic hip examinations if there are indications to do so, such as abnormal values recorded during physical therapy assessments or hip pain. The legal caregivers give oral consent and, if possible, the children give oral assent prior to participation in CPUP. Participation can be discontinued at any time, and the decision to withdraw will not affect the healthcare received. The Regional Ethical Review Board in Lund, Sweden (443-99, revised 2009), approved the study.

2.2 Measures

Age was analysed as a continuous variable and sex as a dichotomous variable. Gross motor function was classified by physical therapists according to the GMFCS Expanded and Revised version, which consists of five levels, where GMFCS level I signifies the highest gross motor functional level and GMFCS level V the most severely restricted. The GMFCS has good validity and reliability5,20,21 and is stable over time.22 Pain in the lower extremities was assessed with four dichotomous items for back, hips, knees and feet (Is it known/believed that the person is in pain?). Pain was self-reported or reported by a caregiver.

Spasticity was measured by physical therapists, and the measures included the Modified Ashworth scale (MAS)23 and scissoring. In this study, MAS scores of 1 or 1+ were combined and coded as 1, corresponding to the original Ashworth scale.24 MAS scores for gastrocnemius, hip adductors and knee flexors for both sides were used. Acceptable interrater reliability has been reported for the MAS.25 In CPUP, scissoring is defined as the legs crossing each other in the frontal plane, while moving or at rest, and is coded as none, mild or pronounced.

Bone/joint complications measures included hip displacement, range of motion (ROM), windswept posture and scoliosis. The degree of hip displacement was measured as the migration percentage (MP) according to Reimer.26 Orthopaedic surgeons or radiologists determined the

Key notes

• Bone/joint complications had the strongest direct pathway with pain in the lower extremities, followed by reduced mobility in children and adolescents with cerebral palsy.
• The pathways between spasticity and pain, and age and pain were not significant.
• Other factors than bone/joint complications, reduced mobility and spasticity must be considered when assessing pain in the lower extremities in individuals with cerebral palsy.
variables. The SEM analysis followed the steps outlined by Kline \cite{Schmidtetal}. Multivariate and bivariate analyses were run to screen the variables to ensure that they met the assumptions for SEM. \cite{Schmidtetal} Univariate measures of central tendencies were calculated for the continuous variable, and proportions were calculated for dichotomous, ordinal and categorical variables. Bivariate correlations among variables were computed using Spearman’s rho. SEM was then used to assess the relationships between spasticity, bone/joint complications, mobility and pain. Confirmatory analysis assessed how well the observed measures reflected the latent constructs of pain, spasticity, bone/joint complications and mobility. A series of factor analyses were performed to assess the fit of the measures. Items with high factor loading (> 0.3) most accurately represent the proposed constructs. Hence, factor loadings had to be > 0.3 to be kept in our final models.

The hypothesised SEM was tested to examine the relationships among constructs. Estimates of path coefficients represent the strength of the path between two variables, and standardised regression coefficients are presented. Squared multiple correlation coefficients ($R^2$) were used to explain the variance of factors on outcomes by the SEM. Age, sex, spasticity, bone/joint complications and mobility were the exogenous variables, and pain in the lower extremity the endogenous variable. Rather than excluding cases with missing values, SEM estimates were calculated using maximum likelihood. The criteria for goodness of fit indices were as follows: incremental fit index >0.90, Tucker-Lewis index 0.90, comparative fit index >0.90, and root means square error of approximation <0.08. \cite{Schmidtetal} IBM SPSS Statistics version 25.0 and the analysis of moment structures (AMOS) software (version 25.0.0) were used.

## RESULTS

### 3.1 Descriptive statistics and correlation coefficients

The descriptive statistics are presented in Tables 1-4, and variables are grouped as basic characteristics of the sample and the three hypothesised exogenous latent constructs, respectively.

The correlation coefficients ranged from 0 (‘number of pain sites’ and ‘hip displacement left’) to 0.91 (‘ability to get up from sitting’ and ‘ability to stand’). To minimise multicollinearity, the variable ‘ability to get up from sitting’ was removed. All other variables had correlations < 0.90.

### 3.2 Testing the measurement models

Four confirmatory factor analyses were run to test the measurement models. Some variables were allowed to correlate based on theoretical considerations. Five variables had loadings <0.3 (Figure 2). Hip displacement right (0.27) and left (0.27) and ROM foot dorsal-flexion right (~0.11) and left (~0.15) did not have sufficient factor loadings and were removed from the bone/joint complications measurement model. In the pain measurement model, back pain was removed due to low factor loading (0.14). Observed measures that were removed are marked in red (Figure 2).
**FIGURE 1** Traffic light coding of range of motion variables according to Gross Motor Function Classification System (GMFCS) level

**CLEAR**
- GMFCS I – II
  - Hip abduction ≥40°
  - Knee Extension ≥0°
  - Foot dorsiflexion ≥10°
- GMFCS III – V
  - Hip abduction ≥30°
  - Knee Extension ≥10°
  - Foot dorsiflexion ≥0°

**CAUTION**
- GMFCS I – II
  - Hip abduction 31° - 39°
  - Knee Extension -1° - -9°
  - Foot dorsiflexion 1° - 9°
- GMFCS III – V
  - Hip abduction 21° - 29°
  - Knee Extension -11° - -19°
  - Foot dorsiflexion -9° - 1°

**ACT**
- GMFCS I – II
  - Hip abduction ≤30°
  - Knee Extension ≤-10°
  - Foot dorsiflexion ≤0°
- GMFCS III – V
  - Hip abduction ≤20°
  - Knee Extension ≤-20°
  - Foot dorsiflexion ≤-10°

**FIGURE 2** Measurement models, spasticity, mobility, bone/joint complications and pain in the lower extremities
After removing the observed variables with low loadings from the a priori model, the first structural model to be analysed is depicted in Figure 3. Fifteen iterations were required to fit the model to the data (degrees of freedom = 259). The $R^2$ for pain in lower extremity was 0.12. All parameter estimates were significant ($P < .05$). The estimates of model fit were incremental fit index = 0.918, Tucker-Lewis index = 0.898, comparative fit index = 0.918, and root means square error of approximation = 0.06.

The measures of fit were close to acceptable for this model, but next we explored additional paths to determine if the model fit could be improved. As all parameter estimates in the model were significant, no paths were removed. Two new paths provided significant improvements to the model fit and were also theoretically relevant; the bone/joint complications and age and the wheelchair use outdoors and FMS 500 metres paths were thus allowed to covary.

The modified model is presented in Figure 4 with standardised estimates and covariances marked in green for those added to the original model. The two paths marked in red (spasticity and pain lower extremities) and (age and pain lower extremities) were the only parameter estimates that were not statistically significant ($P > .05$). The $R^2$ for pain in the lower extremities was 0.15.

The modified model required 17 iterations to fit the model to the data (degrees of freedom = 257). The estimates of model fit
TABLE 2  Manifest variables spasticity

|                      | n, (%)   |
|----------------------|----------|
| Scissoring at rest   |          |
| None                 | 2791 (85.7) |
| Mild                 | 211 (6.5)  |
| Pronounced           | 67 (2.1)   |
| Scissoring at movement|          |
| None                 | 2448 (75.2) |
| Mild                 | 394 (12.1)  |
| Pronounced           | 188 (5.8)   |
| Modified Ashworth scale |        |
| Hip adductors        |          |
| 0                    | 1933 (59.4)  |
| 1                    | 698 (21.4)   |
| 2                    | 68 (2.1)     |
| 3                    | 11 (0.3)     |
| Knee flexors         |          |
| 0                    | 1608 (49.4)  |
| 1                    | 1021 (31.4)  |
| 2                    | 254 (7.8)    |
| 3                    | 66 (2.0)     |
| 4                    | 8 (0.2)      |
| Gastrocnemius        |          |
| 0                    | 909 (27.9)   |
| 1                    | 1385 (42.5)  |
| 2                    | 448 (13.8)   |
| 3                    | 195 (6.0)    |
| 4                    | 29 (0.9)     |

were incremental fit index = 0.938, Tucker-Lewis index = 0.921, comparative fit Index = 0.938, and root means square error of approximation = 0.052.

**TABLE 3**  Manifest variables bone/joint complications

|                      | n, (%)   |
|----------------------|----------|
| Hip displacement (Reimer's index), mean (SD) |          |
| Right                | 18 (14.69) |
| Left                 | 18 (14.61) |
| Range of motion      |          |
| Hip abduction        |          |
| Red                  | 467 (14.3) |
| Yellow               | 292 (9.0)  |
| Green                | 1962 (60.3) |
| Knee extension       |          |
| Red                  | 271 (8.3)  |
| Yellow               | 163 (5.0)  |
| Green                | 2612 (80.2) |
| Foot dorsal-flexion  |          |
| Red                  | 426 (13.1) |
| Yellow               | 326 (10.0) |
| Green                | 2288 (70.3) |
| Windswept posture    |          |
| Yes                  | 232 (7.7)  |
| No                   | 2784 (92.3) |
| Scoliosis            |          |
| None                 | 2284 (70.1) |
| Mild                 | 469 (14.4) |
| Moderate             | 119 (3.7)  |
| Severe               | 85 (2.6)   |

4 | DISCUSSION

In this study, we investigated how spasticity, bone/joint complications and mobility were related to pain in the lower extremities in relation to age. Of the three latent constructs, bone/joint complications had the strongest direct pathway with pain in the lower extremities, followed by reduced mobility. The direct pathway between spasticity and pain in the lower extremities was not significant. It is possible, however, that spasticity has an indirect effect on pain through reduced mobility and through bone/joint complications.

In much of the literature on pain in the context of CP, pain increases significantly with age. In the first model we tested, this was supported. However, in the modified model, the direct pathway from age to pain in the lower extremities was no longer significant. This was most likely due to the correlation between age and bone/joint complications. Previous research has shown that muscle contractures in the lower extremities, development of scoliosis and windswept posture increase with age. The development of contracture of the calf muscle, seen as reduced dorsiflexion of the foot, mainly develops before 6 years of age, whereas contractures of the hips and knees mostly develop after 6 years of age. This is in line with the observed findings in the current study. Hip displacement, as measured by Reimer’s index, did not have a high enough loading on the construct of bone/joint complications to be included in the final model. The risk of developing hip displacement is highest among children 3 to 5 years of age. Most children in CPUP showing signs of hip displacement are treated, which means that although hip displacements are indeed painful, the prevalence of hip displacement in children in Sweden today is low, and as such does not contribute substantially to pain in the lower extremities. It is also possible that any effect of hip displacement on pain might be indirect rather than being part of a construct of bone/joint complications. For instance, hip displacement can result in limited hip abduction or windswept...
TABLE 4  Manifest variables mobility

|                              | n, (%)          |
|------------------------------|-----------------|
| Functional mobility scale 5 m (house) |                |
| 1 = wheelchair               | 773 (23.8)a     |
| 2 = walkers/frames           | 132 (4.1)       |
| 3 = crutches                 | 4 (0.1)         |
| 4 = canes (one or two)       | 31 (1.0)        |
| 5 = independent on level surfaces | 527 (16.2)     |
| 6 = independent on all surfaces | 1417 (43.5)    |
| 7 = crawls                   | 204 (6.3)       |
| Functional mobility scale 50 m |                |
| 1 = wheelchair               | 989 (30.4)b     |
| 2 = walkers/frames           | 167 (5.1)       |
| 3 = crutches                 | 2 (0.1)         |
| 4 = canes (one or two)       | 49 (1.5)        |
| 5 = independent on level surfaces | 527 (16.2)     |
| 6 = independent on all surfaces | 1336 (41.0)    |
| Functional mobility scale 500 m |               |
| 1 = wheelchair               | 1069 (32.8)     |
| 2 = walkers/frames           | 74 (2.3)        |
| 3 = crutches                 | 1 (0)           |
| 4 = canes (one or two)       | 34 (1.0)        |
| 5 = independent on level surfaces | 454 (13.9)     |
| 6 = independent on all surfaces | 1267 (38.9)    |
| 7 = cannot complete the distance | 204 (6.3)      |
| Wheelchair use outdoors      |                |
| No                           | 1792 (55.0)     |
| Yes                          | 1252 (38.5)     |
| Able to stand                |                |
| No                           | 113 (3.5)       |
| Yes                          | 2995 (91.9)c    |
| Able to get up from sitting |                |
| No                           | 563 (17.3)      |
| Yes, with help               | 610 (18.7)      |
| Yes, without help            | 1913 (58.8)     |
| Able to use stairs (walk, scoot, crawl) |           |
| No                           | 1115 (34.2)     |
| Yes                          | 1964 (60.3)     |

*131 participants who were coded as ‘cannot complete the distance’ were recoded as wheelchair.
*147 participants who were coded as ‘cannot complete the distance’ were recoded as wheelchair.
*978 were able to stand with support. GMFCS levels IV and V = 773 were able to stand with help or support.

posture, which, in turn, can be more highly associated with pain in the lower extremities.

One of the aims of CPUP is early detection and treatment of secondary complications in the musculoskeletal system, such as contractures, windswept posture and hip displacement. This is done to optimise gross motor function. The relationship shown between pain and bone/joint complications suggests that this preventive approach might reduce some of the pain in the lower extremities in children with CP. In a previous study, the prevalence of pain in Sweden was lower than in many other international studies. One explanation for this may be the preventive work in the CPUP. Another explanation might be differences in methodologies across studies. Nevertheless, the final model explained 15 per cent of the variance of pain in the lower extremities, meaning that there are numerous other factors associated with pain in addition to the ones included herein. To further reduce the pain in this population, research is needed to understand what these additional factors might be. Prevention of secondary conditions such as pain is often only possible at an individual level. In Sweden, virtually all children and adolescents with CP diagnoses have been identified, and the infrastructure and multidisciplinary collaboration is already established through CPUP. If the causes of pain can be targeted more specifically, it might be possible to address pain at a population-based level.

There were a number of limitations in our study. In terms of affecting participation and quality of life, severity and duration of pain might be more relevant than simply studying the presence of pain. Information regarding severity of pain was not included in CPUP until 2018 and could therefore not be included in the analyses. Furthermore, cross-sectional research only provides a snapshot of a phenomenon; longitudinal data analyses would have provided richer data on the pain experience of children with CP. Registry-based research tends to include large samples. In the case of this study, the vast majority of all children and adolescents with CP in Sweden were included. This allows for well-powered studies that are less prone to selection bias than clinic-based studies or pain studies based on convenience samples. Large samples free from selection bias are needed to develop and test models where multiple statistical relationships are tested simultaneously, as in this study. However, registry as well as large-scale survey studies are often limited in terms of the level of detail included. CPUP is a comprehensive multidisciplinary programme that does not focus specifically on pain. While it might be tempting to include more data collection on pain, the clinical reality makes it impractical. A balance has to be struck because longer clinical visits increase the burden on participating families and stress the resources of clinics [19]. Clinicians are of course encouraged to follow-up any pain problems, even though the data are not entered into the CPUP database.

Pain is subjective and cannot be measured objectively, which means that self-report is preferred. In young children, in the case of those who cannot communicate themselves, and at times in the context of severe intellectual disability, proxy-report is required. Mixing both self- and proxy-reports is not ideal. Because we did not want to only include data from those who could self-report, the decision was made to also include proxy-reported data. The results have to be interpreted with that in mind.
Bone/joint complications (ROM, windswept posture and scoliosis) and reduced mobility had the strongest direct relationship with pain in the lower extremities. The variance in pain accounted for in the model was relatively low, indicating that additional factors are associated with pain in the lower extremities in children with CP. Alternatively, the low variance explained could be because the factors associated with pain differ between, for instance, pain site and sex, and this will be investigated in future studies.

CONFLICT OF INTEREST

The authors have no competing interests to declare.

REFERENCES

1. Westbom L, Hägglund H, 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;5(7):41.
2. Yeargin-Allsopp M, Van Naarden BK, Doernberg NS, Benedict RE, Kirby RS, Durkin MS. Prevalence of cerebral palsy in 8-year old children in three areas of the United States in 2002: a multisite collaboration. Pediatrics. 2008;121:547–554.
3. Surveillance of Cerebral Palsy in Europe. Surveillance of cerebral palsy in Europe: a collaboration of cerebral palsy surveys and registries. Dev Med Child Neurol. 2000;42:816–824.
4. Surveillance of Cerebral Palsy in Europe. Prevalence and characteristics of children with cerebral palsy in Europe. Dev Med Child Neurol. 2002;44:633–640.
5. Palisano R, Rosenbaum P, Walter S, Russell D, Wood E, Galuppi B. Development and reliability of a system to classify gross motor function in children with cerebral palsy. Dev Med Child Neurol. 1997;39:214–223.
6. Rosenbaum P, Eliaison A, Hidecker M, Palisano RJ. Classification in childhood disability: focusing on function on the 21st century. J Child Neurol. 2014;29:1036–1045.
7. Frisch D, Msall ME. Health, functioning, and participation of adolescents and adults with cerebral palsy: A review of outcomes research. Dev Disabil Res Revs. 2013;18:84–94.
8. Liptak GS, Accardo PJ. Health and social outcomes of children with cerebral palsy. J Pediatr. 2004;145:536–541.
9. Jahnsen R, Villien L, Aamot G, Stanghelle JK, Holm I. Musculoskeletal pain in adults with cerebral palsy compared with the general population. J Rehab Med. 2004;36:78-84.
10. Van der Sout W, Niebvenhuijsen C, Van den Berg-Emons R, et al. Chronic pain, fatigue, and depressive symptoms in adults with spastic bilateral cerebral palsy. Dev Med Child Neurol. 2012;54:836-842.
11. Sandström K, Alinder J, Öberg B. Descriptions of functioning and health relations to a gross motor classification in adults with cerebral palsy. Disabil Rehabil. 2004;26:1023-1031.
12. Turk MA, Scandale J, Rosenbaum PF, Weber RJ. The health of women with cerebral palsy. Phys Med Rehabil Clin N Am. 2001;12:153-168.
13. Blackman JA, Conaway MR. Adolescents with cerebral palsy: Transitioning to adult healthcare services. Clin Pediatr (Phila). 2014;53:356-363.
14. 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;98:434-440.
15. Alriksson-Schmidt A, Hägglund G. Pain in children and adolescents with cerebral palsy: a population-based registry study. Acta Paediatr. 2016;105:665-670.
16. McKinnon CT, Meehan EM, Harvey AR, Antolovich GC, Morgan PE. Prevalence and characteristics of pain in children and young adults with cerebral palsy: a systematic review. Dev Med Child Neurol. 2019;61:305-314.
17. Ramstad K, Jahnsen R, Skjeldal OH, Diseth TH. Characteristics of recurrent musculoskeletal pain in children with cerebral palsy aged 8 to 18 years. Dev Med Child Neurol. 2011;53:1013-1018.
18. Robb JE, Hägglund G. Hip surveillance and management of the displaced hip in cerebral palsy. J Child Orthop. 2013;7:407-413.
19. Alriksson-Schmidt AI, Arner M, Westbom L, et al. A combined surveillance program and quality register improves management of childhood disability. Disabil Rehabil. 2017;39:830-836.
20. Wood E, Rosenbaum P. The Gross Motor Function. Classification System for cerebral palsy: a study of reliability and stability over time. Dev Med Child Neurol. 2000;42:292-296.
21. Palisano RJ, Rosenbaum P, Bartlett D, Livingston MH. Content validity of the expanded and revised Gross Motor Function Classification System. Dev Med Child Neurol. 2008;50:744-750.
22. Alriksson-Schmidt AI, Nordmark E, Czuba T, Westbom L. Stability of the Gross Motor Function Classification System in children and adolescents with cerebral palsy: a retrospective cohort registry study. Dev Med Child Neurol. 2017;59:641-646.
23. Bohannon RW, Smith MB. Interrater reliability of a Modified Ashworth Scale of muscle spasticity. Phys Ther. 1987;67:206-207.
24. Ashworth B. Preliminary trial of carisoprodol in multiple sclerosis. Practitioner. 1964;192:540-542.
25. Meseguer-Henarejos AB, Sanchez-Meca J, Lopez-Pina JA, Carles-Hernandez R. Inter- and intra-rater reliability of the Modified ashworth scale: a systematic review and meta-analysis. Eur J Phys Rehabil Med. 2018;54:576-590.
26. Reimers J. The stability of the hip in children. A radiological study of the results of muscle surgery in cerebral palsy. Acta Orthop Scand Suppl. 1980;184:1-100.
27. Young NL, Wright JG, Lam P, Rajaratnam K, Stephens D, Wedge JH. Windswept hip deformity in spastic quadriplegic cerebral palsy. Pediatr Phys Ther. 1998;10:94-100.
28. McWhirk LB, Glanzman AM. Within-session inter-rater reliability of goniometric measures in patients with spastic cerebral palsy. Pediatr Phys Ther. 2006;18:262-265.
29. Persson-Bunke M, Czuba T, Hägglund G, Rodby-Bousquet E. Psychometric evaluation of spinal assessment methods to screen for scoliosis in children and adolescents with cerebral palsy. BMC Musculoskelet Disord. 2015;16:351.
30. Graham HK, Harvey A, Rodda J, Nattrass GR, Pirpiris M. The functional mobility scale (FMS). Journal of Pediatrics Orthopedics. 2004;24:514-520.
31. Kline RB. Principles and Practice of Structural Equation Modeling, Third Edition (Methodology in the Social Sciences) (3rd edn). New York, NY: The Guilford Press; 2010.
32. Schumacker RE, Lomax RG. Model fit. In Schumacker RE, Lomax R. A beginner’s guide to structural equation modeling (2nd edn). London, UK: Lawrence Erlbaum Associates, Publishers; 2004.
33. Nordmark E, Hägglund G, Lauge-Pedersen H, Wagner P, Westbom L. Development of lower limb range of motion from early childhood to adolescence in cerebral palsy - a population based study. BMC Med. 2009;7:65.
34. Hägglund G, Pettersson K, Czuba T, Persson-Bunke R-B. Incidence of scoliosis in cerebral palsy. Acta Orthop. 2018;89:443-447.
35. Hägglund G, Lauge-Pedersen H, Persson-Bunke M, Rodby BE. Windswept hip deformity in children with cerebral palsy - a 20-year population based prospective follow-up. J Child Orthop. 2016;10:275-279.
36. Hägglund G, Lauge-Pedersen H, Wagner P. Characteristics of children with hip displacement in cerebral palsy. BMC Musculoskeletal Disord. 2007;26(8):101.

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