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REGULAR ARTICLE

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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 R^2 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 R^2 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

1 | 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]

Abbreviations: AMOS, Analysis of moment structures; CFI, Comparative fit index; CP, Cerebral palsy; CPUP, Cerebral Palsy Follow-up Program; FMS, Functional Mobility Scale; GMFCS, Gross Motor Function Classification Scale; IFI, Incremental fit index; MAS, Modified Ashworth scale; MP, Migration percentage; RMSEA, Root means square error of approximation; ROM, Range of motion; SEM, Structural equation model; TLI, Tucker-Lewis Index.

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,[7,9-12] 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.

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.

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.[19] 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 reliability [5,20,21] 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

MP on anteroposterior pelvic radiographs. MP is defined as the proportion of the femoral head positioned lateral to the acetabular margin and ranges from 0 (fully inside the acetabular margin) to 100 (hip dislocated). Bilateral ROM for hip abduction, knee extension and foot dorsal-flexion (knee extended) was measured by physical therapists in a standardised position according to a manual (www.cpup.se). The ROM values were rounded to the closest multiple of five. CPUP uses the traffic light principle for ROM scores: a green value means *clear*, a yellow value means *caution*, and a red value means *act*. The scores that constitute green, yellow and red values differ by GMFCS level and depend on which ROM site is measured (Figure 1).

Windswept posture was defined as a minimum of 50 per cent difference in ROM between left and right hip in any one of three measurements: abduction, internal and/or external rotation,[27] and it was coded as yes or no. Intraclass correlation coefficients for ROM in the hips in individuals with CP have been reported at 0.58-0.93.[28] To be coded as windswept posture, the difference in ROM between both sides had to be ≥ 15 degrees. Scoliosis was coded as not present, mild, moderate or severe. Scoliosis was determined by clinical examinations by physical therapists and included assessments of the spine in three positions: sitting, upright and forward bending. Scoliosis is graded as mild (discrete curve visible only on thorough physical examination in forward bending), moderate (obvious curve in both upright and forward bending) or severe (pronounced curve preventing upright position without external support). This standardised clinical spinal assessment has shown high interrater reliability, sensitivity, specificity and criterion-related validity compared with radiographic Cobb angle measurement.[29]

Mobility measures included the Functional Mobility Scale (FMS)[30] at 5- (at home), 50- (at school) and 500-metres (in the wider community), respectively. FMS was constructed as a performance measure to classify functional mobility in children while taking into account the range of assistive devices a child might use.[30]

2.3 | Data considerations and statistical analysis

Univariate analyses were used to describe the sample. Structural equation modelling (SEM) was used to evaluate the relationship among four latent constructs: pain in lower extremities, spasticity, mobility and bone/joint complications. In addition, two observed variables were included: age to determine if the experience of pain differed with increasing age and sex to adjust for known differences in the experience of pain in this population. SEM allows for the simultaneous consideration of multiple pathways between the latent variables. The SEM analysis followed the steps outlined by Kline[31]: (a) model specification (measurement and structural models), (b) estimation of free parameters, (c) assessment of fit, (d) model modification and (e) interpretation.

The initial model specification was developed a priori. The measurement model (Figure 2) included four latent variables, and the structural model (Figure 3) included the latent variables and the observed variables age and sex. Several models were specified based on the theoretical model. Univariate and bivariate analyses were run to screen the variables to ensure that they met the assumptions for SEM.[31] 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.[32] IBM SPSS Statistics version 25.0 and the analysis of moment structures (AMOS) software (version 25.0.0) were used.

3 | 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).

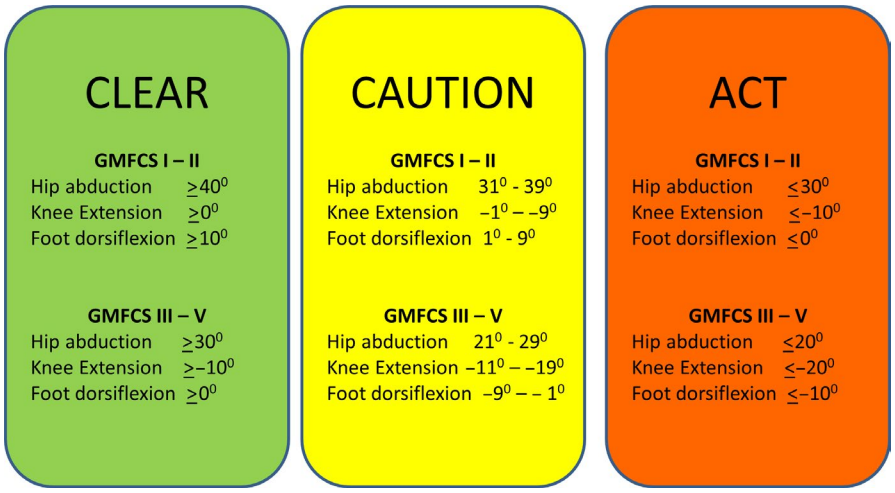

FIGURE 1 Traffic light coding of range of motion variables according to Gross Motor Function Classification System (GMFCS) level

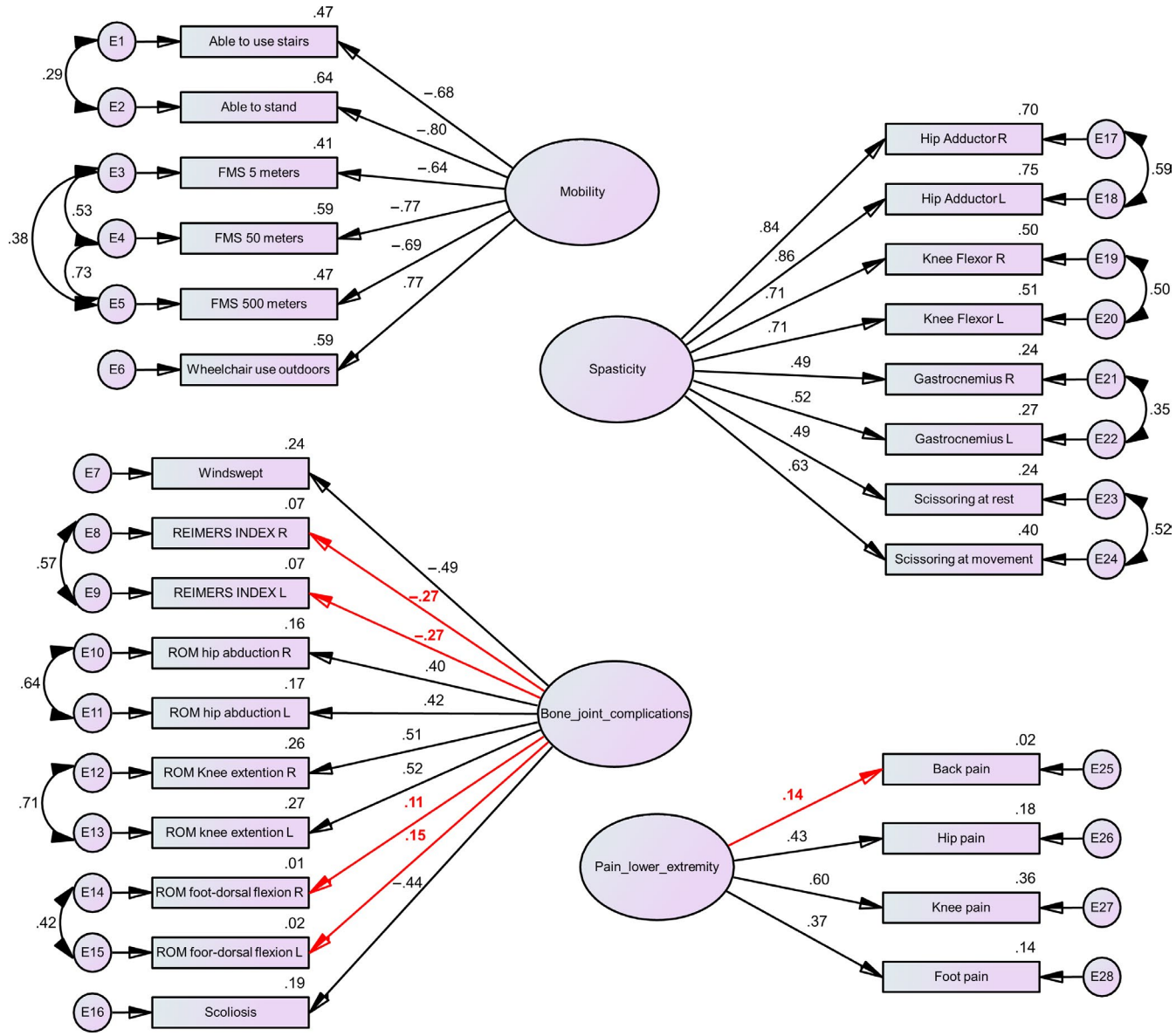

FIGURE 2 Measurement models, spasticity, mobility, bone/joint complications and pain in the lower extremities

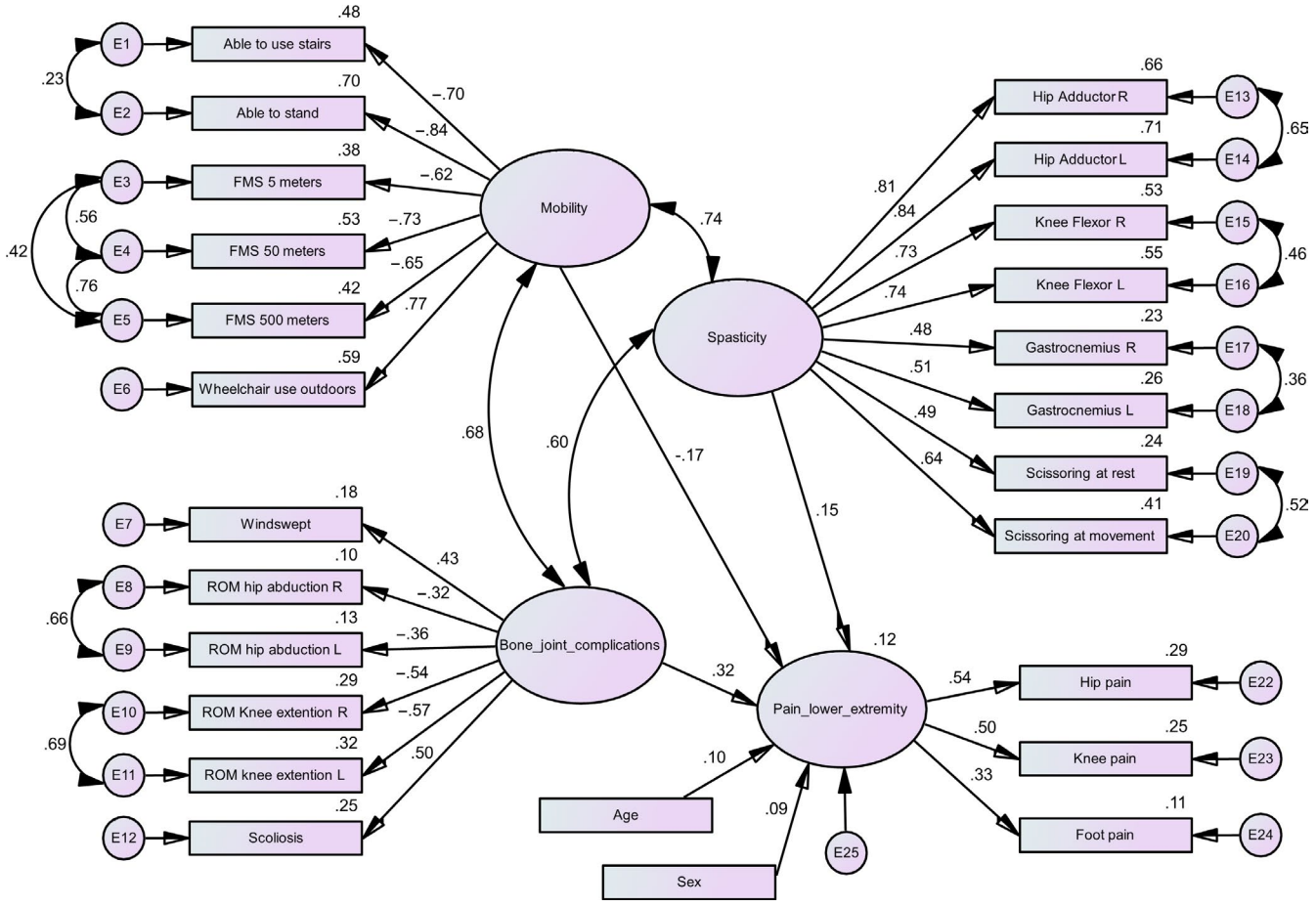

FIGURE 3 Initial structural equation model

TABLE 1 Basic characteristics of sample

	n, (%)
Sex	
Girls	1395 (42.8)
Boys	1861 (57.2)
Gross Motor Function Classification System level	
I	1513 (46.5)
II	550 (16.9)
III	271 (8.3)
IV	469 (14.4)
V	453 (13.9)
Age	
Median years (SD)	9 (3.76)
Pain	
No	2031 (62.4)
Yes	1225 (37.6)
Pain site	
Back	157 (4.8)
Hips	221 (6.8)
Knee	281 (8.6)
Feet	405 (12.4)

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)

	n, (%)	
Modified Ashworth scale	**Right**	**Left**
Hip adductors		
0	1933 (59.4)	1918 (58.9)
1	698 (21.4)	677 (20.8)
2	205 (6.3)	214 (6.6)
3	68 (2.1)	75 (2.3)
4	11 (0.3)	11 (0.3)
Knee flexors		
0	1608 (49.4)	1619 (49.7)
1	1021 (31.4)	983 (30.2)
2	254 (7.8)	260 (8.0)
3	66 (2.0)	69 (2.1)
4	8 (0.2)	5 (0.2)
Gastrocnemius		
0	909 (27.9)	958 (29.4)
1	1385 (42.5)	1353 (41.6)
2	448 (13.8)	421 (12.9)
3	195 (6.0)	180 (5.5)
4	29 (0.9)	31 (1.0)

TABLE 3 Manifest variables bone/joint complications

	n, (%)	
	Right	**Left**
Hip displacement (Reimer's index), mean (SD)	18 (14.69)	18 (14.61)
Range of motion		
Hip abduction		
Red	467 (14.3)	430 (13.2)
Yellow	292 (9.0)	284 (8.7)
Green	1962 (60.3)	1997 (61.3)
Knee extension		
Red	271 (8.3)	264 (8.1)
Yellow	163 (5.0)	157 (4.8)
Green	2612 (80.2)	2605 (80.0)
Foot dorsal-flexion		
Red	426 (13.1)	354 (10.9)
Yellow	326 (10.0)	317 (9.7)
Green	2288 (70.3)	2339 (71.8)
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)	

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.

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.[15] 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,[33] development of scoliosis[34] and windswept posture[35] 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.[33] 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.[36] 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)

[a]131 participants who were coded as 'cannot complete the distance' were recoded as wheelchair.
[b]147 participants who were coded as 'cannot complete the distance' were recoded as wheelchair.
[c]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.[15] 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.

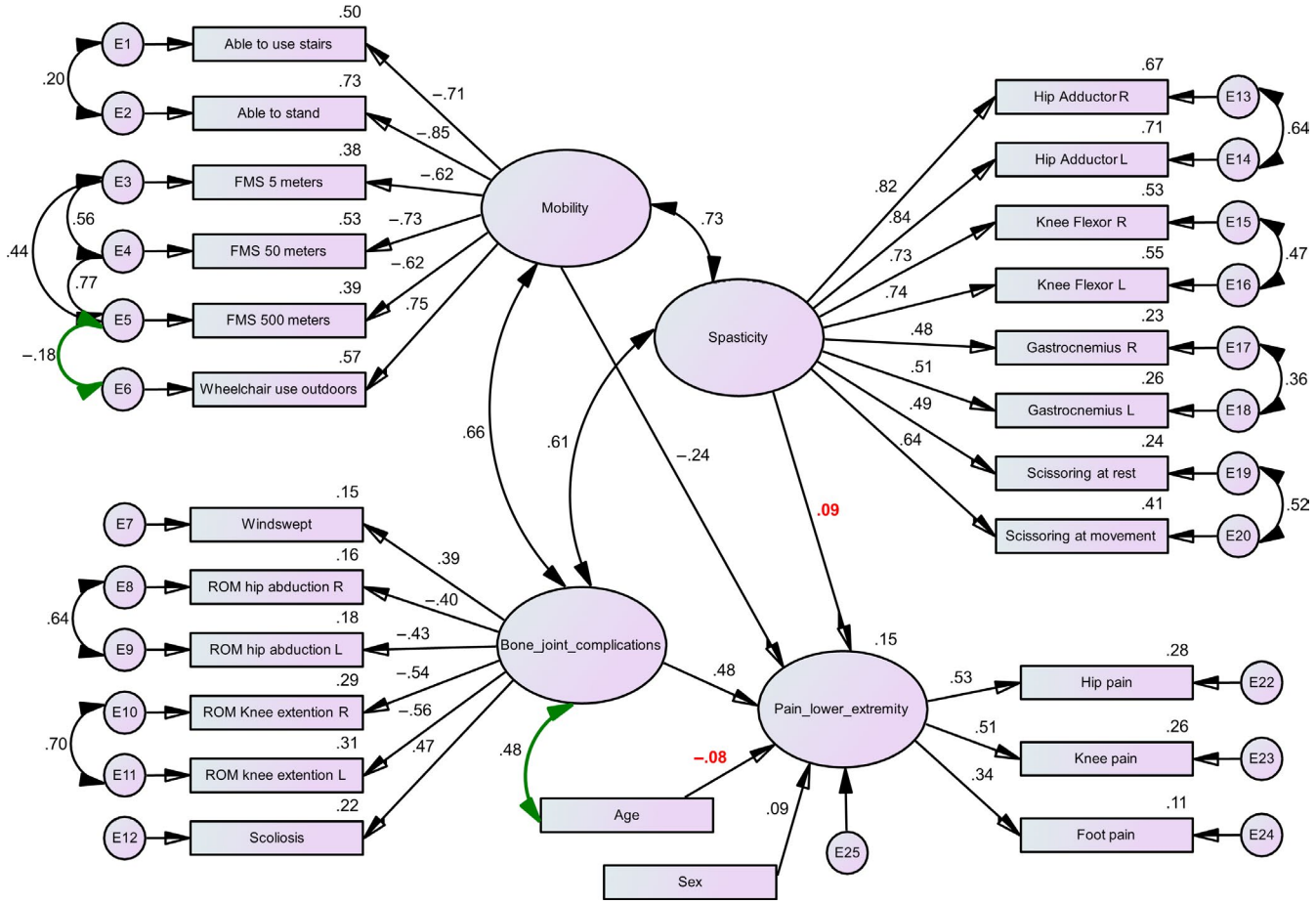

FIGURE 4 Modified structural equation model

5 | CONCLUSIONS

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.

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