Inter- and intra-rater reliability of the head-shaft angle in children with cerebral palsy

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Abstract

Purpose Children with cerebral palsy (CP) are at increased risk for hip dislocation. This can be prevented in most cases using surveillance programmes that include radiographic examinations. Known risk factors for hip dislocation include young age, high Gross Motor Function Classification System (GMFCS) level and high migration percentage (MP). The head-shaft angle (HSA) has recently been described as an additional risk factor. The study aim was to determine inter- and intra-rater reliability of the HSA in a surveillance programme for children with CP.

Methods We included hip radiographs from the CP surveillance programme CPUP in southern Sweden during the first half of 2016. Fifty radiographs were included from children at GMFCS levels II-V, with a mean age of 6.6 (SD 3.2) years. Three raters measured the HSA of one hip (left or right) at baseline and four weeks later; intraclass correlation coefficient (ICC) was used to estimate inter- and intra-rater reliability.

Results Inter- and intra-rater reliability were excellent for the HSA, with ICC 0.92 (95% CI 0.87-0.96) and ICC 0.99 (95% CI 0.98-0.99), respectively.

Conclusion The HSA showed excellent inter- and intra-rater reliability for children with CP, providing further evidence for use of the HSA as an additional factor for identifying risk for further hip displacement or dislocation.

Keywords: Hip; cerebral palsy; head-shaft angle; reliability; hip displacement; CPUP; CPUP hip score

Introduction

Cerebral palsy (CP) is the most common cause of motor disability in children, with 2-3 per 1000 children affected. Their risk for hip dislocation is increased and often occurs at an early age. Hip dislocation may cause pain while lying, sitting, standing and/or walking, and is associated with windswept deformity, pelvic obliquity, contractures and scoliosis. Children with CP should be followed with hip radiographs from an early age to facilitate preventive treatment for those at risk of hip dislocation.

Coxa valga is often seen in CP and can be measured as the neck-shaft angle (NSA), the angle between the femoral neck and shaft. The femoral head is often in valgus and these combined deformities can be measured as the head-shaft angle (HSA) or the angle formed between the perpendicular line between the physis and the femoral shaft (Fig. 1). Forohaar et al showed that the NSA is sensitive to rotation while the HSA only has a 5° measurement error for rotation up to 45°. Therefore, the HSA might be a more useful clinical tool for evaluating the risk of hip dislocation. Well-known risk factors for hip displacement include young age, high migration percentage (MP), more severe Gross Motor Function Classification System (GMFCS) level and a high HSA, all of which have been individually shown to affect the risk for hip displacement. The CPUP Hip Score is a tool for calculating the risk for hip displacement risk (MP >40%) within five years based on age, GMFCS, MP and HSA.

The purpose of this study was to evaluate the HSA inter- and intra-rater reliability in children with CP on radiographs from different radiographic departments in a surveillance programme.

Patients and methods

CPUP, a surveillance programme and national registry for children with CP, was initiated in 1994. Almost all children in Sweden with CP (> 95%) are included in the CPUP. The CPUP programme enrols children with suspected and possible CP as early as possible, in most cases before the age of two years; diagnosis is confirmed at the age of four years by a neuropaediatrician. As part of the CPUP, the

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hips are examined clinically and radiographically using standardised anteroposterior (AP) radiographs, according to a schedule based on the child’s age and GMFCS level (Fig. 2). GMFCS levels I-V are used, in part, to classify CP severity, with a higher level indicating a more severe gross motor impairment.

We included radiographs from children followed in the CPUP from southern Sweden (Skåne and Blekinge) who were examined with an AP pelvic radiograph during the first half of 2016. In total, 107 children were identified and 50 children were finally included (Fig. 3). Radiographs in which < 3 cm of the femoral shaft was visible, measured from the distal part of the lesser trochanter, were excluded (n = 34). We also excluded radiographs in which the physis of the femoral neck was closed (n = 4) and those with previous skeletal hip surgery (n = 12). Several radiographs were also excluded on the basis of both shortness of visible femur and previous skeletal hip surgery (n = 6). One child moved out of the area and was excluded because the collection of her radiograph was impossible (n = 1). A total of 50 patients met the inclusion criteria (Fig. 3).

One hip (left or right) was randomly designated for measurement on each radiograph using the Bernoulli distribution. The HSA was measured independently on all radiographs by three raters on two occasions (two orthopaedic surgeons (GH, JR) and one MD (MH)). Prior to making study measurements, the raters met for a methodological training session at which they developed an agreement on the HSA measurement technique standardisation using radiographs that were not a part of this study. All raters used the same type of ruler (M+R 30 cm plastic), protractor (M+R 10 cm plastic) and a 0.5 mm pencil (MILAN PL1 Look Mechanical Pencil 0.5 mm). Radiographs were printed from the same printer on the same quality paper and all radiographs were measured by hand, anonymously, blinded to others and following the same order (1 to 50) at both baseline and after four weeks. Results were recorded by an independent physiotherapist (ERB). Thereafter, each participant’s age, gender and GMFCS level were recorded.

Southwick et al. described how to measure the HSA in 1967: the first line is drawn through the physis, connecting the inferior and superior margins of the epiphysis; the second line is drawn perpendicular to this line; the third line is drawn through the midshaft of the femur. The HSA is the angle formed between the perpendicular line to the femoral head and the femoral shaft (A).
between the perpendicular line of the femoral head and the femoral shaft (Fig. 1).

The study was approved by the Medical Research Ethics Committee of Lund University (LU-443-99).

Statistical analyses

A Bernoulli distribution was used to randomly select whether the right or left hip was to be measured. The outcome of interest was the HSA measurement in the right or left hip. Inter- and intra-rater reliability were evaluated using the intraclass correlation coefficient (ICC) and 95% confidence interval (95% CI) with two-way random and absolute agreement for single measures.20 ICC for inter-rater reliability was based on the measurements by each of the three raters at the first assessment. ICC for intra-rater reliability was evaluated for all three raters separately and also calculated as an average of all three raters, where the HSA for each child at the first assessment was compared with the HSA at the second assessment. SPSS Statistic version 24 for Windows Software package was used for statistical analyses.

Results

Radiographs of 50 children (25 girls, 25 boys) at GMFCS levels II (n = 10), III (n = 12), IV (n = 15) and V (n = 13) were included in the study. Their mean age was 6.6 (SD 3.2) years (Table 1). Inter-rater reliability for the HSA was excellent, with ICC 0.92 (95% CI 0.87-0.95). Intra-rater reliability for the HSA was also excellent with ICC 0.98, 0.94 and 0.98 for each of the respective raters (Table 2). ICC showed excellent values (ICC 0.99) when intra-rater
**Table 1. Characteristics of the 50 children included and degrees of their HSA by the three raters at the first and second measurements.**

| Case | Age | Gender | GMFCS | Hip | HSA measurement 1 | HSA measurement 2 |
|------|-----|--------|-------|-----|-------------------|-------------------|
|      |     |        |       |     | Rater A | 151 | 157 | 155 | 150 | 154 | 154 |
| 1    | 8   | M      | 4     | R   |           |      |      |      |      |      |      |
| 2    | 5   | M      | 3     | L   | 154      | 162 | 160 | 153 | 162 | 160 |      |
| 3    | 4   | F      | 3     | R   | 161      | 160 | 153 | 157 | 155 | 155 |      |
| 4    | 12  | M      | 5     | R   | 156      | 162 | 160 | 155 | 158 | 157 |      |
| 5    | 12  | F      | 3     | L   | 170      | 167 | 164 | 173 | 172 | 167 |      |
| 6    | 3   | F      | 4     | L   | 178      | 175 | 173 | 174 | 178 | 175 |      |
| 7    | 13  | F      | 5     | R   | 156      | 160 | 151 | 155 | 160 | 148 |      |
| 8    | 4   | F      | 2     | R   | 151      | 154 | 155 | 148 | 157 | 154 |      |
| 9    | 3   | F      | 5     | R   | 155      | 157 | 162 | 153 | 161 | 162 |      |
| 10   | 8   | F      | 5     | R   | 173      | 170 | 177 | 170 | 172 | 175 |      |
| 11   | 8   | M      | 2     | R   | 162      | 160 | 159 | 161 | 169 | 160 |      |
| 12   | 8   | M      | 5     | R   | 174      | 175 | 172 | 174 | 175 | 176 |      |
| 13   | 6   | F      | 4     | L   | 171      | 173 | 173 | 170 | 176 | 173 |      |
| 14   | 6   | M      | 5     | R   | 159      | 160 | 162 | 160 | 164 | 164 |      |
| 15   | 3   | M      | 4     | R   | 192      | 191 | 192 | 193 | 193 | 192 |      |
| 16   | 4   | F      | 2     | L   | 168      | 167 | 168 | 168 | 169 | 166 |      |
| 17   | 5   | F      | 3     | L   | 167      | 169 | 171 | 166 | 169 | 171 |      |
| 18   | 10  | M      | 4     | L   | 147      | 148 | 150 | 147 | 150 | 149 |      |
| 19   | 6   | M      | 4     | L   | 166      | 170 | 170 | 166 | 167 | 169 |      |
| 20   | 4   | M      | 2     | R   | 148      | 153 | 154 | 149 | 157 | 152 |      |
| 21   | 8   | F      | 2     | R   | 160      | 152 | 143 | 158 | 150 | 145 |      |
| 22   | 7   | M      | 2     | R   | 150      | 151 | 148 | 147 | 152 | 150 |      |
| 23   | 7   | F      | 2     | L   | 156      | 156 | 158 | 152 | 160 | 158 |      |
| 24   | 6   | F      | 4     | L   | 177      | 173 | 175 | 174 | 176 | 178 |      |
| 25   | 5   | M      | 3     | R   | 167      | 165 | 165 | 168 | 170 | 164 |      |
| 26   | 10  | F      | 3     | L   | 164      | 162 | 159 | 162 | 161 | 161 |      |
| 27   | 10  | F      | 5     | L   | 170      | 169 | 169 | 168 | 173 | 168 |      |
| 28   | 8   | F      | 3     | R   | 166      | 163 | 164 | 163 | 164 | 163 |      |
| 29   | 7   | M      | 4     | L   | 160      | 161 | 160 | 161 | 161 | 160 |      |
| 30   | 7   | M      | 2     | R   | 161      | 159 | 160 | 159 | 163 | 157 |      |
| 31   | 5   | M      | 4     | L   | 172      | 170 | 170 | 169 | 172 | 172 |      |
| 32   | 4   | F      | 4     | L   | 168      | 170 | 167 | 166 | 171 | 167 |      |
| 33   | 3   | M      | 5     | R   | 159      | 161 | 161 | 157 | 162 | 159 |      |
| 34   | 1   | F      | 5     | L   | 169      | 172 | 173 | 171 | 176 | 171 |      |
| 35   | 13  | M      | 5     | L   | 169      | 169 | 168 | 169 | 168 | 171 |      |
| 36   | 8   | F      | 4     | R   | 172      | 177 | 179 | 168 | 172 | 179 |      |
| 37   | 8   | M      | 4     | R   | 170      | 168 | 168 | 167 | 170 | 168 |      |
| 38   | 6   | M      | 3     | L   | 164      | 168 | 170 | 163 | 163 | 170 |      |
| 39   | 6   | F      | 3     | L   | 148      | 155 | 155 | 151 | 153 | 154 |      |
| 40   | 6   | M      | 4     | R   | 162      | 159 | 159 | 158 | 159 | 161 |      |
| 41   | 6   | F      | 2     | R   | 163      | 164 | 164 | 165 | 164 | 163 |      |
| 42   | 6   | M      | 5     | R   | 178      | 180 | 178 | 179 | 179 | 176 |      |
| 43   | 5   | F      | 3     | L   | 168      | 169 | 172 | 168 | 171 | 173 |      |
| 44   | 1   | F      | 3     | R   | 158      | 162 | 162 | 157 | 164 | 163 |      |
| 45   | 1   | M      | 5     | L   | 164      | 166 | 172 | 159 | 176 | 173 |      |
| 46   | 8   | F      | 4     | L   | 146      | 152 | 156 | 146 | 154 | 151 |      |
| 47   | 7   | M      | 2     | L   | 163      | 167 | 166 | 165 | 168 | 164 |      |
| 48   | 14  | M      | 3     | L   | 140      | 142 | 137 | 137 | 140 | 137 |      |
| 49   | 12  | F      | 4     | L   | 154      | 155 | 149 | 155 | 153 | 154 |      |
| 50   | 1   | M      | 5     | L   | 165      | 167 | 165 | 165 | 165 | 164 |      |

M, male; F, female; GMFCS, Gross Motor Function Classification System (I-V); R, right; L, left; HSA, head-shaft angle (°)
reliability was calculated using averaged values of the three raters. ICCs for both intra- and inter-rater reliability were statistically significant ($p < 0.001$).

Discussion

The HSA showed excellent inter- and intra-rater reliability for assessing children with CP. In relation to the GMFCS level distribution among the total population, there are many radiographs of children at GMFCS level II in this sample. This is because we excluded children with previous skeletal surgery, which is more common at higher GMFCS levels. A study strength was that the entire population of children with CP in one region (Skåne/Blekinge) were included, comprising 11 radiology departments, which is representative of the CP surveillance programme and allows us to generalise these results to other populations.

Our high inter-rater reliability values are consistent with those of Foroohar et al., who also showed excellent inter-rater reliability for the HSA (ICC 0.94) when measured by three raters for 39 children (70 hips) with CP, and a mean age of 7.6 years. Slightly lower inter-rater reliability for the HSA (ICC 0.79) was reported by Lee et al., when two raters measured 384 children (384 hips) aged 3 to 17 years. That there were only two raters and the children were slightly older might explain these lower inter-rater reliability values.

Intra-rater reliability for a single observer has been previously evaluated by Chougule et al., by whom the HSA was measured in 100 children (350 radiographs), aged 8.8 (3 to 18) years, with CP at GMFCS levels III-V. Follow-up of at least five years was included and radiographs from 103 children typically developed served as controls. Intra-observer reliability of 0.88 for the HSA for this single observer was estimated with Lin’s concordance correlation coefficient; our results were higher, with excellent intra-rater reliability for all three raters (0.94 to 0.98) despite working in different settings.

To measure the HSA correctly, it is important that the radiograph is of sufficient quality. It is desirable to have at least 3 cm of femur distal from trochanter minor visible to allow a correct measurement of the diaphyseal axis. In some hips, the physis shape can make knowing where to draw the line challenging. Some examples displaying how we decided to draw the lines that form the HSA are shown in Figure 4. Radiographs where the physis were closed were excluded in this study. If the physeal line is still visible, it is possible to measure the HSA also after physeal closure. However, hip displacement in CP usually occurs before the age of ten years and measuring HSA at time of skeletal maturity is seldom of clinical importance. In this study, only one hip was measured on each radiograph to eliminate the risk of the other hip being a confounder as they are not independent. High reliability indicates that the HSA is suitable for hip surveillance programmes.

Table 2. Inter- and intra-rater reliability for the three raters measuring the HSA.

| Reliability       | ICC  | 95% CI    | p-value |
|-------------------|------|-----------|---------|
| Inter-rater       | 0.92 | 0.87      | 0.96    | < 0.001 |
| Intra-rater       | 0.99 | 0.98      | 0.99    | < 0.001 |
| Rater A           | 0.98 | 0.94      | 0.99    | < 0.001 |
| Rater B           | 0.94 | 0.88      | 0.96    | < 0.001 |
| Rater C           | 0.98 | 0.97      | 0.99    | < 0.001 |

ICC, intraclass correlation coefficient; HSA, head-shaft angle.

**Fig. 4** Four radiograph examples for how lines were decided to be drawn to measure the head-shaft angle (HSA). The radiographs show: (a) undulating physis: a line is drawn parallel to the epiphysis to connect the metaphysis by the lateral and medial margins; (b) round physis: a line is drawn at the base of the epiphysis, connecting the superior and inferior margins; (c) oval physis: a line is drawn through the midline of the physis; (d) rounded epiphysis: a line is drawn parallel to the proximal end of the metaphysis, since the epiphysis is rounded and does not have distinct superior and inferior margins. In all cases (a-d) a second line is drawn perpendicular to the first line and connected with a third line through the midshaft femur to form the HSA.
In a bone model, Foroohar et al.\textsuperscript{12} showed that the HSA is not sensitive to femoral rotation up to 45°, with a measurement error of 5° or less. This might explain why reliability in the present study was high even though radiographs were from different radiology departments with the one instruction that radiographs should be anteroposterior. Foroohar et al.\textsuperscript{12} stated that the proximal physis was not well demarcated in some of the radiographs, which might have caused their lower inter-rater reliability. Lee et al.\textsuperscript{9} did not describe how much of the femoral shaft was visible on the radiographs, which might also explain their lower ICC. Further, their mean age was 9.1 years; it may have been more difficult to measure the HSA of older children due to their less distinct growth plates.

Van der List et al.\textsuperscript{22} measured the HSA of both hips on radiographs from 50 children (GMFCS levels II-V) at ages two, four and seven years. They concluded that the HSA and the GMFCS level in two-year-olds were greater in hips that will displace. In another study, van der List et al.\textsuperscript{23} compared reference HSA values in 50 children (both hips) with CP at GMFCS levels II-V with the contralateral hip in 33 children (one hip) with developmental dysplasia of the hip. In the normally developed hip, HSA decreased by 2° per year. In children with CP and GMFCS II and III, the HSA decreased by 0.6° and 0.9° per year, respectively. The change in HSA was not statistically significant in children at GMFCS levels IV and V.

Chougule et al.\textsuperscript{21} found no correlation between the HSA and hip migration. The children in their study were older (8.8 years) than in our previous studies (3.5 years),\textsuperscript{14,15} which might explain the lack of significant correlation; risk for hip displacement is proportionally higher at younger ages.

We conclude that the HSA shows excellent intra- and inter-rater reliability for children with CP and can be a useful assessment tool for predicting hip displacement in CP.

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Ethical Statement

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

The study was approved by the Medical Research Ethics Committee of Lund University (LU-443-99).

Informed consent

Informed consent was obtained from all individual participants included in the study.

References

1. Hagberg B, Hagberg G, Beckung E, Uvebrant P. Changing panorama of cerebral palsy in Sweden. VIII. Prevalence and origin in the birth year period 1991–94. Acta Paediatr 2001;90:271–277.
2. Nordmark E, Hägglund G, Lagergren J. Cerebral palsy in southern Sweden I. Prevalence and clinical features. Acta Paediatr 2001;90:1271–1276.
3. Hägglund G, Laude-Pedersen H, Wagner P. Characteristics of children with hip displacement in cerebral palsy. BMC Musculoskelet Disord 2007;8:101.
4. Brunner R, Baumann JU. Clinical benefit of reconstruction of dislocated or subluxated hip joints in patients with spastic cerebral palsy. J Pediatr Orthop 1994;14:290–294.
5. Hägglund G, Laude-Pedersen H, Persson Bunke M, Rodby-Bousquet E. Windswept hip deformity in children with cerebral palsy: a population-based prospective follow-up. J Child Orthop 2016;10:275–279.
6. Persson-Bunke M, Hägglund G, Laude-Pedersen H. Windswept hip deformity in children with cerebral palsy. J Pediatr Orthop B 2006;15:335–338.
7. Persson-Bunke M, Hägglund G, Laude-Pedersen H, Wagner P, Westbom L. Scoliosis in a total population of children with cerebral palsy. Spine (Phila Pa 1976) 2002;37:E708–E713.
8. Hägglund G, Alriksson-Schmidt A, Laude-Pedersen H, et al. Prevention of dislocation of the hip in children with cerebral palsy: 20-year results of a population-based prevention programme. Bone Joint J 2014;96-B:1546–1552.
9. Lee KM, Kang JY, Chung CY, et al. Clinical relevance of valgus deformity of proximal femur in cerebral palsy. J Pediatr Orthop 2010;30:720–725.
10. Bobroff ED, Chambers HG, Sartoris DJ, Wyatt MP, Sutherland DH. Femoral anteversion and neck-shaft angle in children with cerebral palsy. Clin Orthop Relat Res 1999;364:194–204.
11. Southwick WO. Osteotomy through the lesser trochanter for slipped capital femoral epiphysis. J Bone Joint Surg Am 1967;49:807–815.
12. Foroohar A, McCarthy JJ, Yucha D, Clarke S, Brey J. Head-shaft angle measurement in children with cerebral palsy. J Pediatr Orthop 2009;29:248–250.
13. 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.
14. Hermanson M, Hägglund G, Riad J, Wagner P. Head-shaft angle is a risk factor for hip displacement in children with cerebral palsy. *Acta Orthop* 2015;86:229-232.

15. Hermanson M, Hägglund G, Riad J, Rodby-Bousquet E, Wagner P. Prediction of hip displacement in children with cerebral palsy: development of the CPUP hip score. *Bone Joint J* 2015;87-B:1441-1444.

16. Hägglund G, Andersson S, Düppe H, et al. Prevention of dislocation of the hip in children with cerebral palsy. The first ten years of a population-based prevention programme. *J Bone Joint Surg (Br)* 2005;87-B:95-101.

17. Hägglund G, Andersson S, Düppe H, et al. Prevention of severe contractures might replace multilevel surgery in cerebral palsy: results of a population-based health care programme and new techniques to reduce spasticity. *J Pediatr Orthop B* 2005;14:269-273.

18. Palisano R, Rosenbaum P, Walter S, et al. Development and reliability of a system to classify gross motor function in children with cerebral palsy. *Dev Med Child Neurol* 1997;39:214-223.

19. 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.

20. McGraw KO, Wong SP. Forming inferences about some intraclass correlations coefficients [Correction]. *Psychol Methods* 1996;1:390.

21. Chougule S, Dabis J, Petrie A, Daly K, Gelfer Y. Is head-shaft angle a valuable continuous risk factor for hip migration in cerebral palsy? *J Child Orthop* 2016;10:651-656.

22. van der List JP, Witbreuk MM, Buizer AI, van der Sluijs JA. The prognostic value of the head-shaft angle on hip displacement in children with cerebral palsy. *J Child Orthop* 2015;9:129-135.

23. van der List JP, Witbreuk MM, Buizer AI, van der Sluijs JA. The head-shaft angle of the hip in early childhood: a comparison of reference values for children with cerebral palsy and normally developing hips. *Bone Joint J* 2015;97-B:1291-1295.