Original Clinical Article

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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.

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Introduction

Cerebral palsy (CP) is the most common cause of motor disability in children, with 2-3 per 1000 children affected.[1,2] Their risk for hip dislocation is increased and often occurs at an early age.[3] Hip dislocation may cause pain while lying, sitting, standing and/or walking,[4] and is associated with windswept deformity, pelvic obliquity, contractures and scoliosis.[5-7] Children with CP should be followed with hip radiographs from an early age to facilitate preventive treatment for those at risk of hip dislocation.[8]

Coxa valga is often seen in CP[9] and can be measured as the neck-shaft angle (NSA),[10] 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).[11] Forohaar et al[12] showed that the NSA is sensitive to rotation while the HSA only has a 5° measurement error for rotation up to 45°.[12] 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),[13] more severe Gross Motor Function Classification System (GMFCS) level and a high HSA,[14] all of which have been individually shown to affect the risk for hip displacement.[15] 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.[15]

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.[8,16,17] Almost all children in Sweden with CP (> 95%) are included in the CPUP.[8] 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

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).[8,16,17] GMFCS levels I-V are used, in part, to classify CP severity, with a higher level indicating a more severe gross motor impairment. [18,19]

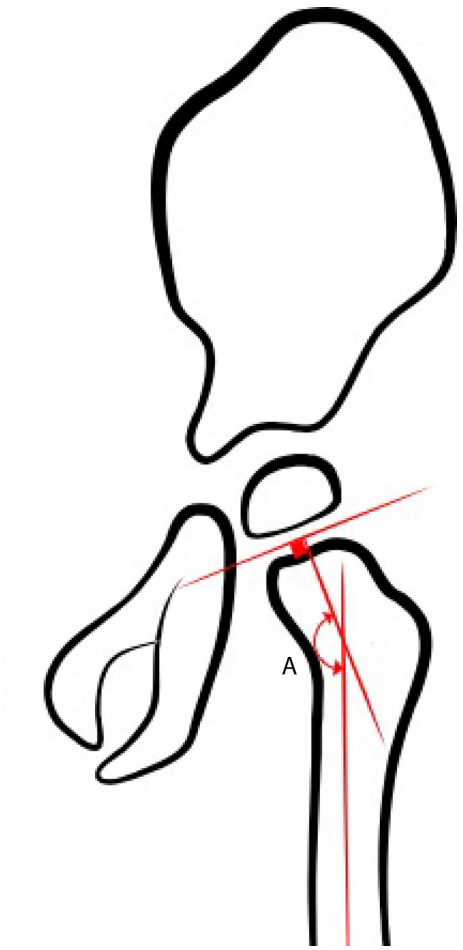

Fig. 1 Measurement of the head-shaft angle (HSA). The first line is drawn through the inferior and superior margins of the epiphysis. The second line is drawn perpendicular to this line. The third line is drawn through the midshaft of the femur. The HSA is the angle formed between the perpendicular line to the femoral head and the femoral shaft (A).

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[11] described how to measure the HSA in 1967: the first line is drawn through the physis, connecting the inferior and the superior margins of the epiphysis; the second line is drawn perpendicular to the first; the third line is drawn through the femur midshaft.[11] In this study, we drew a line midway through the femur through two dots, approximately 1 and 3 cm distal from the lesser trochanter, respectively. The HSA is the angle formed

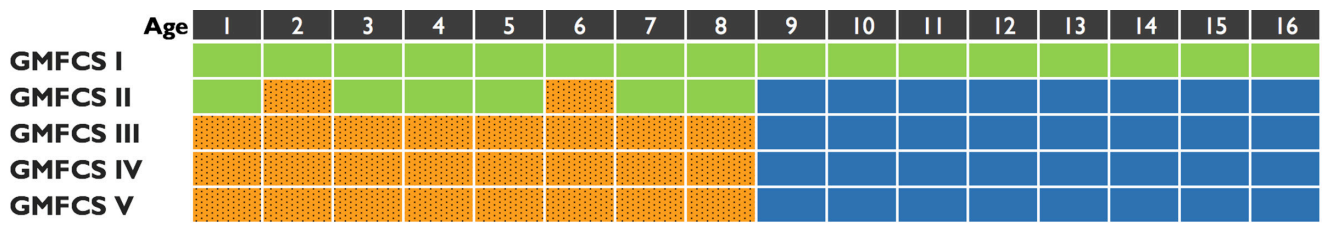

Fig.2 Schedule for radiographic hip examination in the CPUP. Green = radiographic examination is considered if clinical examination and migration percentage indicate risk for hip dislocation, otherwise no radiograph is performed. Orange = radiographic examination should be performed at this age and GMFCS level, which for GMFCS levels III-V is once a year until the age of eight years and thereafter individually. Blue = follow-up with radiographic examination individually, depending on earlier radiographs and clinical examination.

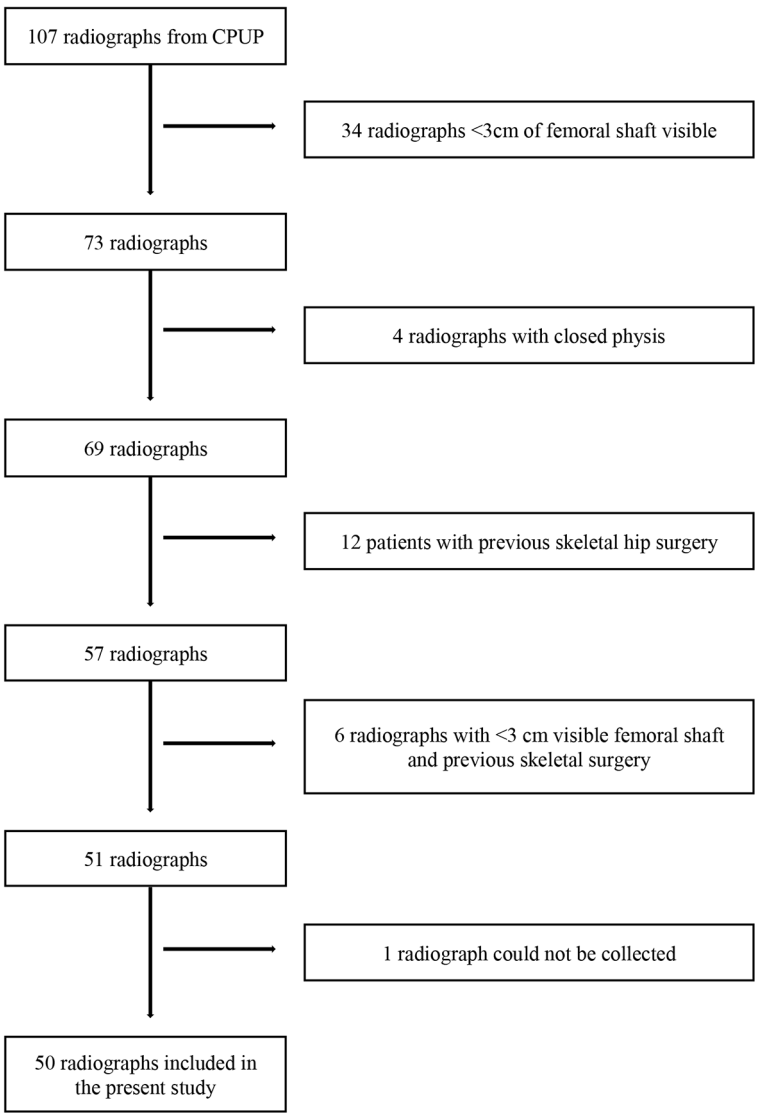

Fig. 3 Flow chart of the study selection process.

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.

					HSA measurement 1			HSA measurement 2		
Case	Age	Gender	GMFCS	Hip	Rater A	Rater B	Rater C	Rater A	Rater B	Rater C
1	8	M	4	R	151	157	155	150	154	154
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,[12] 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,[9] 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,[21] 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[3] 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.

		95% CI		
	ICC	Lower bound	Upper bound	p-value
Inter-rater reliability	0.92	0.87	0.96	< 0.001
Intra-rater reliability	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

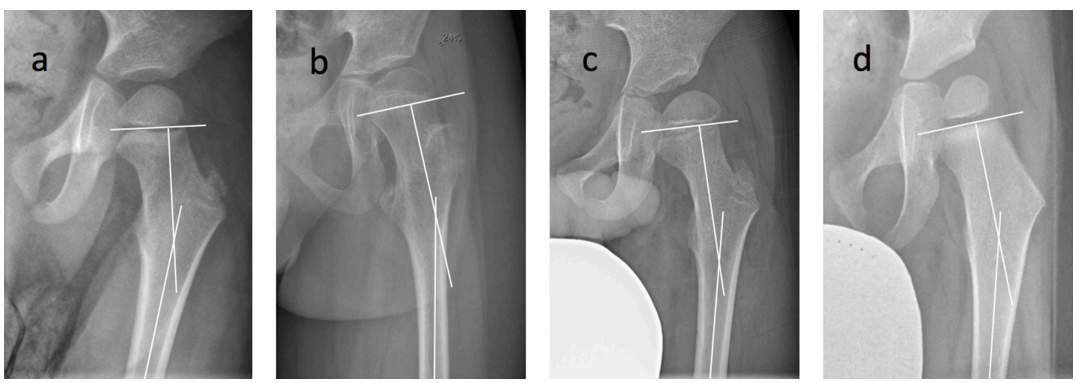

Fig. 4 Four radiograph examples for how lines were decided to be drawn to measure the head-shaft angle (HSA). The radiographs show: (**a**) undulating physis: a line is drawn parallel to the epiphysis to connect the metaphysis by the lateral and medial margins; (**b**) round physis: a line is drawn at 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[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[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[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[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[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[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), [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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COMPLIANCE WITH ETHICAL STANDARDS

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ETHICAL STATEMENT

Ethical approval

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.

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