Proximal femoral geometry in cerebral palsy
A POPULATION-BASED CROSS-SECTIONAL STUDY

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There is much debate about the nature and extent of deformities in the proximal femur in children with cerebral palsy. Most authorities accept that increased femoral anteversion is common, but its incidence, severity and clinical significance are less clear. Coxa valga is more controversial and many authorities state that it is a radiological artefact rather than a true deformity.

We measured femoral anteversion clinically and the neck-shaft angle radiologically in 292 children with cerebral palsy. This represented 78% of a large, population-based cohort of children with cerebral palsy which included all motor types, topographical distributions and functional levels as determined by the gross motor function classification system.

The mean femoral neck anteversion was 36.5° (11° to 67.5°) and the mean neck-shaft angle 147.5° (130° to 178°). These were both increased compared with values in normally developing children. The mean femoral neck anteversion was 30.4° (11° to 50°) at gross motor function classification system level I, 35.5° (8° to 65°) at level II and then plateaued at approximately 40.0° (25° to 67.5°) at levels III, IV and V. The mean neck-shaft angle increased in a step-wise manner from 135.9° (130° to 145°) at level I to 163.0° (151° to 178°) at level V. The migration percentage increased in a similar pattern and was closely related to femoral deformity.

Based on these findings we believe that displacement of the hip in patients with cerebral palsy can be explained mainly by the abnormal shape of the proximal femur, as a result of delayed walking, limited walking or inability to walk. This has clinical implications for the management of hip displacement in children with cerebral palsy.

Cerebral palsy is the most common cause of childhood physical disability in developed countries with an incidence of 2 per 1000 live births.1 It is a static encephalopathy which may result in progressive musculoskeletal pathology,1,2 the key features of which are contractions of muscle-tendon units, bony torsional deformity and instability of joints.3 Hip displacement is the second most common musculoskeletal deformity after equinus, affecting over one-third of children with cerebral palsy.4 The incidence is directly related to the child’s functional level as determined by the gross motor function classification system (GMFCS).4

Subluxation of the hip in cerebral palsy may contribute to an inefficient gait in those children who can walk, and cause pain and difficulty in nursing care and perineal hygiene in those who cannot.5-10

The two most important anatomical factors defining the proximal femur are anteversion of the femoral neck in the transverse plane and the femoral neck-shaft angle in the coronal plane. There are many synonyms for both. Femoral neck anteversion has been referred to as either femoral antetorsion, medial femoral torsion, internal femoral torsion or persistent fetal alignment.11 By definition, femoral neck anteversion is the anterior torsion of the femoral neck relative to the femoral condyles in the transverse plane.12 It does not necessarily occur in the femoral neck and may be seen at any point along the length of the femur.12 The neck-shaft angle has been referred to as femoral inclination and the centrum-collum diaphseal angle of Müller.13 When increased above the normal range it is termed coxa valga and when decreased below the normal range it is termed coxa vara.

The geometry of the proximal femur may be a factor in displacement of the hip in children with cerebral palsy and may cause gait problems in the absence of subluxation.14 The three-dimensional (3D) nature of the proximal femur can be difficult to quantify and may require clinical and radiological
assessments. Clinical examination using the trochanteric prominence angle test is a reliable technique for measuring femoral neck anteversion and is useful as a clinical screening tool.\textsuperscript{15,17} Axial imaging using CT or MRI is considered by some to be the method of choice for measurement of femoral neck anteversion.\textsuperscript{15} However, CT exposes children to relatively high doses of radiation and access to MRI may be limited.\textsuperscript{15} Their use in a large population-based study may not be ethical or practical. Measurement of the neck-shaft angle from neutral anteroposterior radiographs of the pelvis is not accurate when there is increased anteversion.\textsuperscript{18} Accurate measurement of the neck-shaft angle from an anteroposterior radiograph may be performed with the hips internally rotated by approximately the same degree of anteversion present.\textsuperscript{18}

Many previous studies have identified increased femoral neck anteversion in children with cerebral palsy.\textsuperscript{13,18,21} This deformity may contribute to intoed gait, problems of gait and instability of the hip.\textsuperscript{2,10,14} Coxa valga in cerebral palsy has been less thoroughly investigated and is more controversial.\textsuperscript{13,18,21} An increased neck-shaft angle has been associated with hip subluxation, but not with dysfunction of gait.\textsuperscript{10,14} The aetiology of both femoral neck anteversion and coxa valga in cerebral palsy has also been debated. Some investigators have attributed femoral neck anteversion to persisting fetal alignment.\textsuperscript{15} Others have suggested that both increased femoral neck anteversion and coxa valga are acquired because of muscle imbalance, spasticity and inability to walk.\textsuperscript{5,10}

Our aim was to measure femoral neck anteversion and the neck-shaft angle in a series of children with cerebral palsy including all motor types, topographical distributions and functional levels. The secondary aim was to investigate the relationship between proximal femoral anatomy, gross motor function and displacement of the hip.

Patients and Methods
This was a retrospective, population-based, cross-sectional study of children who were identified from a statewide cerebral palsy register, which has a high degree of accuracy and case ascertainment.\textsuperscript{22} Approval was obtained from our institutional review board. We have followed children born between 1990 and 1992 at our institution. Approximately one-third have required hip surgery. We included children who did and who did not have hip surgery. In order to ensure that the effects of this surgery would not bias the results of the cohort under investigation, all the data were taken from clinical and radiological assessments before hip surgery. No child had been treated with intrathecal baclofen pumps or by selective dorsal rhizotomy. Some had been treated by injection of Botulinum neurotoxin A into the adductor muscles. However, this was not thought to have changed the structure of the proximal femur and has had a minimal effect on the incidence of hip displacement.\textsuperscript{23}

Cerebral palsy register: demographic data and the gross motor function classification system. Information obtained from the register included the child’s date of birth, motor type, topographical distribution and gross motor function according to the classification system. GMFCS levels were also obtained from records from the gait laboratory or hip surveillance clinic and cross-checked for discrepancies which were then resolved by discussion with the child’s community or hospital-based physiotherapist and developmental paediatrician. The GMFCS is a valid and reliable tool for the classification of gross motor function\textsuperscript{24} and has been described for various age bands.\textsuperscript{24,25}

Clinical measurements of femoral neck anteversion by the trochanteric prominence angle test. Routine clinical and biomechanical evaluations were performed in the gait laboratory for walking children (GMFCS I to III) and in hip surveillance clinics for non-walking children (GMFCS IV and V). As part of the standardised physical examination, measurement of femoral neck anteversion by the trochanteric prominence angle test\textsuperscript{17} was performed by two experienced physiotherapists. One examined the child and stabilised the leg in position while the other used a goniometer to record measurements.

With the child lying prone, the knee was flexed to 90° so that the tibia was vertical and perpendicular to the couch. The prominence of the greater trochanter was palpated at the same time as the hip was rotated internally and externally by the first physiotherapist. When the prominence of the greater trochanter was palpated most laterally, the femoral neck was thought to be parallel to the plane of the couch (in the coronal plane). At this point, a second physiotherapist measured the deviation of the tibia away from the true vertical with a standardised long-arm goniometer to determine the anteversion (Fig. 1). The direct reading (\(\alpha\)) was subtracted from the vertical reference point of 90°. Therefore femoral neck anteversion = 90° - \(\alpha\). As this was part of routine scheduled examinations, measurements were made only once. Internal rotation of the hip at the
point when the greater trochanter was palpated most laterally signified anteversion. External rotation when the greater trochanter was palpated most laterally indicated retroversion. This technique has been shown to have high inter-rater\textsuperscript{17,26-28} and very high intra-rater reliability.\textsuperscript{16,27,28}

The trochanteric prominence angle test is relatively easy to learn and to perform.\textsuperscript{17}

Measurement of the neck-shaft angle and migration percentage. Children had routine radiographs performed on the hip during hip surveillance or before femoral derotation osteotomy as part of surgery to improve gait. Most were standard anteroposterior radiographs. However, an inclusion criterion for our study was that each child had at least one internal hip rotation anteroposterior view of the proximal femur performed from which the true neck-shaft angle could be measured (Figs 2 and 3). These radiographs were obtained with the child in the supine position and with the hips internally rotated approximately 30\degree to 40\degree. This amount of internal rotation fell within the range for accurate measurements of the neck-shaft angle, when there was concomitant increased femoral neck anteversion.\textsuperscript{29}

The measurement of the migration percentage also followed a strictly defined protocol with good reliability and validity.\textsuperscript{30-33} Migration percentage was measured from the last pre-operative neutral anteroposterior pelvic radiograph. After drawing a Hilgenreiner\textsuperscript{34} (horizontal) and Perkins\textsuperscript{35} (perpendicular) line on the radiograph, the migration percentage was calculated as the percentage of the ossified femoral head that remained lateral to the Perkins line\textsuperscript{30} (Fig. 4).

In summary, the inclusion criteria for the study were:

Registration on the statewide cerebral palsy register before commencement of the study and a birth date between January 1, 1990 and December 31, 1992; survival to the age of six years; well-documented clinical records detailing the motor type, topographical distribution and gross motor function classification system level for the ages of six to 12 years; anteroposterior radiographs of the pelvis of good quality taken in the neutral position and internal hip rotation; measurement of femoral neck anteversion by the trochanteric prominence angle test.

The exclusion criterion was the absence of one or more of these criteria.

A total of 374 children met the inclusion criteria. Nine without hip subluxation died before the age of six years and were excluded from the study. A further 16 were lost to follow-up. Radiological documentation with an internal rotation view was not available in 57 children leaving 292 (78\%) in the study. The mean age at examination was 9.3 years (3 to 15). Table I gives details according to motor type and topographical distribution.

Statistical analysis. This was performed by a medical statistician (KS). For each child, the mean femoral neck anteversion, neck-shaft angle and migration percentage values were calculated by averaging the relevant right and left hip measurements. Normally distributed outcomes were summarised by mean and SD and analysed by linear regression. Non-normally distributed outcomes were summarised by median and interquartile range and analysed by linear regression using bootstrapped SEMs. For
each skewed outcome 2000 bootstrap samples were taken, and bias-corrected accelerated confidence intervals (CIs) were calculated. The mean femoral neck anteverision, neck-shaft angle and migration percentage measurements were regressed separately on indicator variables representing gross motor functional classification system categories. The Wald test was used to calculate an overall p-value for the effect of the gross motor functional classification system level for each regression. All analyses were performed using Stata version 9.2 (Stata Corp LP, College Station, Texas).

Results

There were 86 children (29%) who were at GMFCS level I. The distribution of children in GMFCS levels II to V was 55 (19%), 41 (14%), 50 (17%) and 60 (21%), respectively.

The mean femoral neck anteverision for the whole cohort was 36.5° (11° to 67.5°) and the mean neck-shaft angle 147.5° (130° to 178°).

Femoral neck anteverision was elevated at all gross motor functional classification system levels compared with that of normally developing children (Table II). There was very strong evidence for an association between the mean femoral neck anteverision and the GMFCS level (p < 0.001, linear regression analysis). Children in level III to V had the highest. The mean femoral neck anteverision increased with levels I to III (30.4°(11° to 50°) 35.5° (8° to 65°) and 40.5° (25° to 67.5°)) and plateaued at this level for levels IV and V (40.1° (25° to 60°) and 40.5° (31° to 55°)). The 95% CIs for levels I, II and III did not overlap, while for levels III, IV and V did overlap (Fig. 5).

The neck-shaft angle was increased at each GMFCS level compared with that in normally developing children (Table II). Children at GMFCS level I had nearly normal neck-shaft angle, while those in level V had a grossly elevated neck-shaft angle. There was very strong evidence for an association between the mean neck-shaft angle and GMFCS level (p < 0.001, bootstrapped linear regression analysis). The mean neck-shaft angle increased in a step-wise manner from GMFCS levels I to V (135.9° (130° to 145°) 141.0° (130° to 145°), 148.8° (138° to 171°), 155° (138 to 165°) and 163° (151° to 178°). There is no overlap between the 95% CI’s for the five levels. (Fig. 6)

There was also very strong evidence for an association between the migration percentage and GMFCS level (p < 0.001, bootstrapped linear regression analysis). As with the neck-shaft angle, the migration percentage increased in a step-wise manner from GMFCS levels I to V (8.1%, 13.0%, 25.0%, 36.8%, 46.2%). There is no overlap between the 95% CI’s for the five levels (Fig. 7). A summary of the overall results is shown in Figure 8.

<table>
<thead>
<tr>
<th>Table I. Distribution according to motor type in the 292 children</th>
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<td>Motor type and distribution</td>
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<tr>
<td>Spastic</td>
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<td>Hemiplegia</td>
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<th>Table II. Details of previous studies comparing normally-developing children with those in the current study</th>
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<td>Imaging</td>
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<tr>
<td>Rogers³⁶</td>
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<tr>
<td>Dunlap et al¹¹</td>
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<td>Shands and Steele¹⁹</td>
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<td>Fabry et al²⁰</td>
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<td>Normal children</td>
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* NR, not recorded
Discussion

Many investigators have studied the development of the proximal femoral geometry from birth to skeletal maturity.11,18,19,36-40 Few anatomical studies have been performed on preserved fetuses from early gestation to term.41,42 The limited literature suggests that femoral neck anteversion increases in utero, especially during the second half of gestation.41-43 It is speculated that this is due to the mechanical stresses in utero.41 Radiological evaluation of femoral neck anteversion has been performed on cohorts of children of varied ages.11,19,20,36-40 Anatomical investigation of femora has also been performed, mostly on adult specimens.44-51 There is a consensus that femoral neck anteversion decreases from a mean of 35° to 40° at birth to approximately 10° to 15° at skeletal maturity.11,19,20,36-42,47-50 This is thought to be due to a combination of genetic influences, intrauterine moulding and the biomechanical environment around the hip produced during standing and walking.41,43,52 The decrease in femoral neck anteversion follows an exponential curve such that most remodelling occurs early in life, decreasing from approximately 40° at birth to 25° at four years of age (Fig. 9).11,19,20,37

The natural history and development of the neck-shaft angle are more controversial and have been less well studied. There have been very few prenatal investigations of the neck-shaft angle, and no studies have demonstrated significant changes in utero.41,42 Radiological studies have mainly used indirect measurement techniques, using mathematical conversion tables to correct the apparent to the true neck-shaft angle.19,38,39 The consensus from the indirect measurement methods suggests a pattern of slowly decreasing neck-shaft angle with age from a mean of 135° to 140° at birth to 125° at skeletal maturity.19,38,39

The question of the true versus apparent neck-shaft angle is especially important when it is increased as it is in early childhood and in children with cerebral palsy. Radiologically, the true neck-shaft angle can only be measured when the hip is internally rotated to the amount of femoral neck anteversion present. Radiography performed in such a manner eliminates the superimposed femoral anteversion, and allows accurate direct measurement of the neck-shaft angle.18 (Figs 2 and 3). Kay et al29 investigated the effects of femoral rotation on the projected neck-shaft angle using a cadaver dry femur and mathematical modelling. The effects of rotation when there was an increased neck-shaft angle and femoral neck anteversion were also explored. As the femoral neck anteversion increased, further internal rotation of the hip was required to maintain accurate measurements of the neck-shaft angle.29 When the femoral neck anteversion was 40°, measurements of the neck-shaft angle remained accurate to 10° if the hip was rotated internally by 75° to 5°.29 When the femoral neck anteversion was in the range of 30° to 40°, accuracy was optimal when the hips were internally rotated approximately 30° to 40°.29

![Bar chart showing that the mean femoral neck anteversion (FNA) increases from gross motor function classification system (GMFCS) levels I to III, and then plateaus at GMFCS levels III to V. Confidence intervals shown on bars.](image1)

![Bar chart showing that the mean neck-shaft angle (NSA) increases in a stepwise fashion from gross motor function classification system (GMFCS) levels I to V. Confidence intervals shown on bars.](image2)

![Bar chart showing that the mean migration percentage (MP) increases in a stepwise fashion from gross motor function classification system (GMFCS) levels I to V. Confidence intervals shown on bars.](image3)
our study, the mean femoral neck anteversion ranged between 30.4° and 40.5°. The neck-shaft angle was measured from radiographs taken in approximately 30° to 40° of internal rotation of the hip. Therefore the methodology used and the accuracy of neck-shaft angle measurements in our study are supported by the findings of Kay et al.29 To date, there have been no studies which have determined the natural history and age-related changes in the neck-shaft angle as measured by the ‘true’ method. The normal decrease in femoral neck anteversion and neck-shaft angle with age is believed to be related to remodelling of the proximal femur due to biomechanical forces during walking and normal muscle activity across the hip.52-54
at these levels and elevated compared with that at levels I and II. Across all five GMFCS levels, there was relatively little difference in the mean femoral neck anteversion which also suggested that increased anteversion is mainly the result of persisting fetal alignment.

The neck-shaft angle data in our study were in agreement with the only other study using direct measurement techniques of true neck-shaft angles.18 Our results showed a stepwise increase in the neck-shaft angle with decreasing functional ability according to GMFCS level. This suggested that coxa valga is an acquired deformity related to functional limitations. Although the neck-shaft angle at GMFCS levels I, II and III (mostly walking children) showed some increase over that in normal children, the clinically important increase was found in children at levels IV and V (non-walking children). However, it must be noted that the CIs were wide about the mean.

In our study, the migration percentage and neck-shaft angle increased stepwise with the GMFCS level. At levels IV and V, the migration percentage reached the radiological threshold for clinically important subluxation (> 30%). This confirmed previous theories that the combination of both a high neck-shaft angle and femoral neck anteversion predisposed to subluxation of the hip.10,15-17

The major strengths of our study were that it was a large, population-based study, incorporating all motor types and severities of cerebral palsy in children, most of whom had standardised clinical and radiological follow-up. The major weakness was that it was cross-sectional rather than longitudinal, and age may have been a significant confounder. Furthermore, approximately one-third of the children in our cohort required hip surgery, which was offered when the migration percentage exceeded 30%. Therefore this was not a longitudinal study of the natural history of the proximal femoral geometry in cerebral palsy. Finally, a clinical measure of femoral neck anteversion was used rather than the current method of choice of axial imaging.15 However, the accuracy of axial imaging for measurement of femoral neck anteversion is controversial, especially where there is true coxa valga.58 The trochanteric prominence angle test is reliable, but its accuracy, especially postoperatively and in overweight individuals is debated.59 Its use has been supported as a screening tool.15-17

In conclusion, our study has confirmed that children with cerebral palsy have both an increased femoral neck anteversion and neck-shaft angle. These deformities are closely related to gross motor function. An increase in femoral neck anteversion may be caused by persistent fetal alignment due to delayed walking. An increased neck-shaft angle is directly related to limitations in gross motor function. Both deformities are related to displacement of the hip. The migration percentage increased with increasing limitation in gross motor function in a stepwise fashion similar to the neck-shaft angle. The combination of an elevated femoral neck anteversion and neck-shaft angle contributed to increased hip displacement.

This information is relevant clinically when planning intervention for hip displacement. Techniques to reduce spasticity have been shown not to reduce or prevent hip displacement in children with cerebral palsy.23,60,61 The effects of adductor lengthening as a prophylaxis against hip displacement are variable, but in the main disappointing.10,62-64 The benefits of bony reconstructive surgery compared with those of adductor surgery have been supported by Barrie and Galasko.63 The severity and prevalence of bony deformity in the proximal femur in children with cerebral palsy reported in our study may add support to this view.

**Supplementary Material**

A table showing a summary of results according to GMFCS is available with the electronic version of this article on our website at www.jbjs.org.uk

No benefits in any form have been received or will be received from a commercial party related directly or indirectly to the subject of this article.

**References**

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