Anatomic Relationship of the Femoral Neurovascular Bundle in Patients With Congenital Femoral Deficiency

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Background: Congenital femoral deficiency (CFD) is a rare condition that affects the morphology of the hip and surrounding soft tissues. Bony deformity and distorted muscular anatomy are well known, but no studies have described the relationship of the femoral neurovascular (NV) bundle to surgically relevant anatomic landmarks. The authors compared the location of the femoral NV bundle on the affected side in patients with CFD with the unaffected side. The authors hypothesized that the bundle on the pathologic side would be in an abnormal position relative to the unaffected side.

Methods: Thirty-three patients diagnosed with unilateral CFD who had undergone preoperative magnetic resonance imaging of the pelvis were included in our study. The authors identified the femoral NV bundle on the axial cuts and measured its distance from the anterior superior iliac spine (ASIS), anterior inferior iliac spine (AIIS), and lesser trochanter (LT). Anatomic percent change and absolute measurements were then compared and correlated with associated boney deformities and the Paley classification.

Results: The distance from the femoral NV bundle to the ASIS, AIIS, and LT was significantly different compared with the unaffected side. The AIIS absolute distance and AIIS percent change significantly correlated with the neck-shaft angle of the proximal femur.

Conclusions: In patients with CFD, the femoral NV bundle seems to be further from the LT and closer to the AIIS on the affected side when compared with the unaffected side. magnetic resonance imaging may be helpful to understand the course of the femoral NV bundle before reconstruction in patients with CFD; however, the authors recommend identification of the femoral NV bundle before transection of the proximal rectus femoris tendon to provide safe surgical care.

Level of Evidence: Level IV—case-control study of diagnostic studies.

Key Words: congenital femoral deficiency, congenital anomalies, MRI, children, femoral neurovascular bundle

(*J Pediatr Orthop* 2020;00:000–000)

Congenital femoral deficiency (CFD) is a preaxial, longitudinal deficiency of the lower extremity that is associated with a variety of bony and muscular deformities.[1] The incidence of CFD is ~1 in 50,000 to 200,000 births; however, the cause of this abnormality remains unknown in most cases.[2–4] Previous reports have discussed the bony deformities (adduction, extension, and external rotation), soft tissue contractures (hip flexion, abduction), and abnormal development of the muscles that contribute to this clinical appearance.[5] Treatments for these deformities vary on the basis of the severity and may include supportive orthotics and prosthetics, joint reconstruction, and/or limb equalization procedures.[6,7] Although anatomic characteristics of CFD have been described in several studies,[5,8] to our knowledge, no previous studies have investigated the relationship of the femoral neurovascular (NV) bundle in CFD to other anatomic structures pertinent to reconstruction. Understanding the location of the NV bundle and its relationship to other structures is extremely important for a safe reconstruction of the proximal femur and acetabulum. The purpose of this study was to compare the location of the femoral NV bundle relative to the anterior superior iliac spine (ASIS), anterior inferior iliac spine (AIIS), and lesser trochanter (LT) on the affected side with the unaffected side in patients with CFD, and to determine if any of the skeletal deformities correlate with the location of the NV bundle. We hypothesized that the NV bundle on the affected side would have an abnormal relationship with the aforementioned landmarks and would correlate with the neck-shaft angle of the femur, acetabular index, and femoral length discrepancy.

METHODS

This study was a retrospective study and was approved by the local institutional review board (#2018-133). The research methods were carried out in accordance with

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A.J.H.: conceptualization. Y.H.K. and A.J.H.: data curation. D.S.F., T. J.R., A.J.H.: formal analysis. Y.H.K.: funding acquisition. A.J.H. and Y.H.K.: investigation. Y.H.K., A.J.H., and T.J.R.: methodology. A.J.H. and Y.H.K.: project administration; writing – original draft. D.P.: resources. A.J.H., D.P., and D.S.F.: supervision. A.J.H. and T.J.R.: validation. A.J.H.: visualization. A.J.H. and D.S.F.: writing review and editing.

This study was supported by a grant from the National Research Foundation of Korea Grant funded by the Korean Government (NRF-2018R1C1B5039286).

The authors declare no conflicts of interest.

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DOI: 10.1097/BPO.0000000000001709

institutional guidelines and regulations. We identified 134 patients between September 2009 and June 2018 that were diagnosed with CFD. The inclusion criteria were as follows: no previous surgical treatment, unilateral CFD, a magnetic resonance imaging (MRI) scan of the pelvis, and radiographs that included the anterior-posterior pelvis and full-length teleroentgenogram. Eighty-seven patients were excluded because they did not have the preoperative MRI scans of the pelvis available for analysis. We excluded an additional 9 patients because of bilateral CFD and 5 patients because of motion artifacts on the MRI scans that distorted the analysis. In total, 33 patients were enrolled in the study. The mean age at the time of the MRI evaluation was 2 years and 6 months (range, 5 mo to 6 y and 10 mo). The patient position in the MRI was supine with the hips and knees in a natural, resting position. We classified each patient using the Paley classification based on the radiographs and MRI scans. The femoral NV bundle, ASIS, AIIS, and LT were identified on the T1 and T2 axial cuts of the MRI scans, and the distance of the bundle from the ASIS, AIIS, and LT was measured (Fig. 1). The distance from the NV bundle was measured from their closest identifiable margins on the selected axial image. The femoral vein was identified by its medial relation in the bundle as a larger lumen with a thinner wall, and the artery was identified as having a smaller lumen and thicker wall.[9] These anatomic measurements were then compared with the same measurements from the contralateral side. Anatomic percent change was calculated and performed to understand how the distance on the affected side compared with the unaffected side. Anatomic percent change is defined as: the distance measured on the CFD side minus the distance measured on the unaffected side, the difference then divided by the distance measured on the unaffected side and finally, the quotient multiplied by 100. The absolute distances and anatomic percent changes were correlated with the associated bony deformities and the assigned Paley classification. The neck-shaft angle and acetabular index were measured on the frontal plane radiographs or MRI scans. Femoral length was measured on the teleroentgenogram. Statistical analyses were performed using GraphPad Prism version 8.00 for Windows (GraphPad Software, La Jolla, CA, www.graphpad.com). The level of significance was set at $P<0.05$. The paired t test was used to compare the measured differences on both the affected and unaffected sides. The Pearson correlation coefficient was used to correlate the distances measured and relative anatomic ratios with the radiographic deformities. The Spearman correlation coefficient was used to compare the distances and percent changes with the acetabular index percent change and the Paley classification.

RESULTS

Thirty-three patients were included in our study, and 19 of the patients were classified as Paley type 1, of which 2 patients were Paley type 1A, 10 patients were Paley type 1B1, and 7 patients were Paley type 1B2. For the

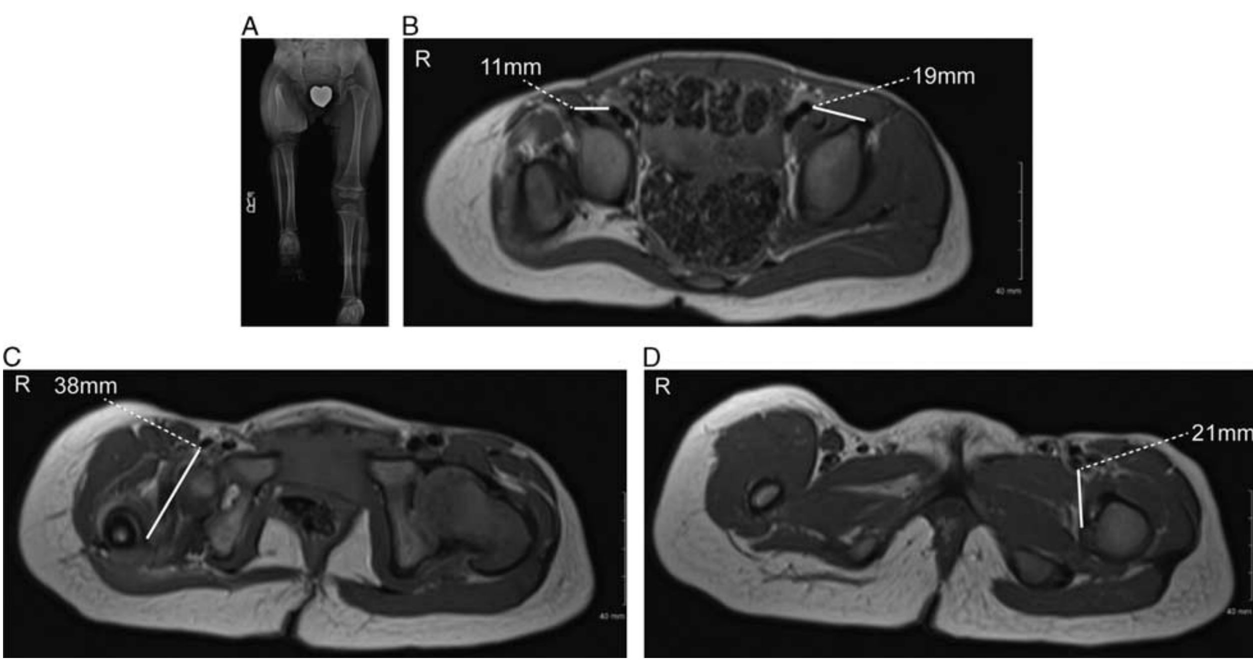

FIGURE 1. Measurement of neurovascular bundle distance from landmarks. A, A 24-month-old girl had Paley 1B2 right congenital femoral deficiency. B, The right femoral neurovascular bundle distance from the anterior inferior iliac spine was 11 mm on the congenital femoral deficiency side, and the left femoral neurovascular bundle distance from the anterior inferior iliac spine was 19 mm on the normal side. C, The right femoral neurovascular bundle distance from the lessor trochanter was 38 mm on the congenital femoral deficiency side. D, The left femoral neurovascular bundle distance from the lessor trochanter was 21 mm on the normal side.

TABLE 1. Distance between Anatomic Landmarks and the Femoral NV Bundle, Radiographic Deformity Measurements and the Anatomic Percent Change

Parameter	CFD Side	Unaffected Side	Percent Change (%)	P
Distance from femoral NV bundle to ASIS (mm)	37 ± 7	39 ± 8	−4 ± 4	< 0.001
Distance from femoral NV bundle to AIIS (mm)	13 ± 3	23 ± 5	−40 ± 15	< 0.001
Distance from femoral NV bundle to LT (mm)	37 ± 8	21 ± 6	88 ± 55	< 0.001
Neck-shaft angle (deg.)	81 ± 30	143 ± 8	−43 ± 22	< 0.001
Acetabular index (deg.)	27 ± 5	20 ± 4	33 ± 30	< 0.001
Femur length (mm)	70 ± 27	184 ± 45	−61 ± 14	< 0.001

AIIS indicates anterior inferior iliac spine; ASIS, anterior superior iliac spine; CFD, congenital femoral deficiency; LT, lesser trochanter; NV, neurovascular.

TABLE 3. Correlation between the Anatomic Percent Change and Deformity of the Femur

	ASIS Percent Change	AIIS Percent Change	Lesser Trochanter Percent Change
Neck-shaft angle	0.045 ($P = 0.809$)	0.798 ($P < 0.0001$)	−0.105 ($P = 0.560$)
Acetabular Index	−0.339 ($P = 0.062$)	−0.485 ($P < 0.01$)	0.043 ($P = 0.812$)
Femoral length percent change	0.280 ($P = 0.128$)	0.068 ($P = 0.709$)	0.114 ($P = 0.529$)
Paley classification	0.049 ($P = 0.795$)	−0.396 ($P < 0.05$)	0.003 ($P = 0.988$)

R/ρ values are listed with their respective significance.
AIIS indicates anterior inferior iliac spine; ASIS, anterior superior iliac spine; LT, lesser trochanter.

remaining 14 patients, the most common type was Paley type 2A. None of the patients reviewed were Paley type 3 or 4. Fifteen patients were female and 18 patients were male individuals. Table 1 summarizes the anatomic absolute distances, the radiographic deformities, and the percent change. The distance comparisons regarding the ASIS, AIIS, and LT were all statistically significant ($P < 0.001$). The correlations describing the relationship of the anatomic absolute distances and the skeletal deformities/classification have been compiled in Table 2 and the correlations describing the relationship of the anatomic percent changes and the skeletal deformities are seen in Table 3. A strong, positive correlation was seen between the AIIS percent change and the neck-shaft angle with a linear regression best fit line described by the equation $y = 0.394x - 72.14$. The Paley classification did not correlate with acetabular index ($\rho = 0.152$, $P = 0.400$) or the femoral length percent change ($\rho = -0.341$, $P = 0.075$). However, the classification did inversely correlate with the neck-shaft angle ($\rho = -0.465$, $P < 0.01$).

DISCUSSION

CFD represents a complex spectrum of osseous, muscular, and ligamentous deformities. Although treatment remains controversial, improvements in imaging and instrumentation, and a better understanding of deformities enable us to perform complex reconstructions and preserve the limb.[6,10–12] Previous studies have utilized MRI to describe different soft tissue features in CFD including muscle size and vessel morphology, but these studies were performed with a small number of patients and described anatomic characteristics without considering their surgical importance.[5,8,13] At our institution, MRI has been used to differentiate between delayed ossification of the femoral neck and a true pseudoarthrosis. To our knowledge, we are not aware of any other studies that use MRI to describe the location of the femoral NV bundle as it relates to anatomic landmarks pertinent in the hip and proximal femoral reconstruction in patients with CFD.

Common boney deformities of the proximal femur in patients with CFD are varus, external rotation, and extension.[12] Soft tissue deformities include abduction, flexion, and external rotation.[12] Proximal femoral and acetabular reconstruction is performed to correct these deformities and prepare the hip for lengthening. One of the structures at risk during the reconstruction of the acetabulum and proximal femur is the femoral NV bundle. Knowledge about the location of the femoral NV bundle is very important especially with distorted anatomy to prevent iatrogenic injuries. Although there are no published cases in the literature of iatrogenic injury during CFD reconstruction, there are anecdotal reports of the femoral nerve crossing the AIIS before rectus femoris transection.

One portion of reconstruction utilizes the Smith-Petersen interval (sartorius/tensor fascia lata) for the release of the rectus femoris and iliopsoas tendon to assist with the soft tissue flexion contracture. The ASIS is the first boney landmark and easily detectable with palpation. In the unaffected child's hip, the ASIS is a safe distance from the femoral NV bundle. In our study, there was a significant difference between the affected (37 mm) and unaffected sides (39 mm). However, we do not believe this to be clinically significant as the mean percent change is 4% and the mean distance remains > 3 cm from this surgical landmark.

The next surgical landmark during the dissection is the AIIS. During the approach, we routinely identify the

TABLE 2. Correlation Between the Absolute Distances and Deformity of the Femur

	Distance to the ASIS	Distance to the AIIS	Distance to the Lesser Trochanter
Neck-shaft angle	−0.425 ($P < 0.05$)	0.458 ($P < 0.01$)	−0.434 ($P < 0.05$)
Acetabular Index	0.429 ($P < 0.05$)	−0.196 ($P = 0.275$)	0.382 ($P < 0.05$)
Femoral length percent change	0.064 ($P = 0.732$)	−0.117 ($P = 0.518$)	−0.412 ($P < 0.05$)
Paley classification	−0.001 ($P = 0.996$)	0.049 ($P = 0.795$)	0.2296 ($P = 0.199$)

R/ρ values are listed with their respective significance.
AIIS indicates anterior inferior iliac spine; ASIS, anterior superior iliac spine.

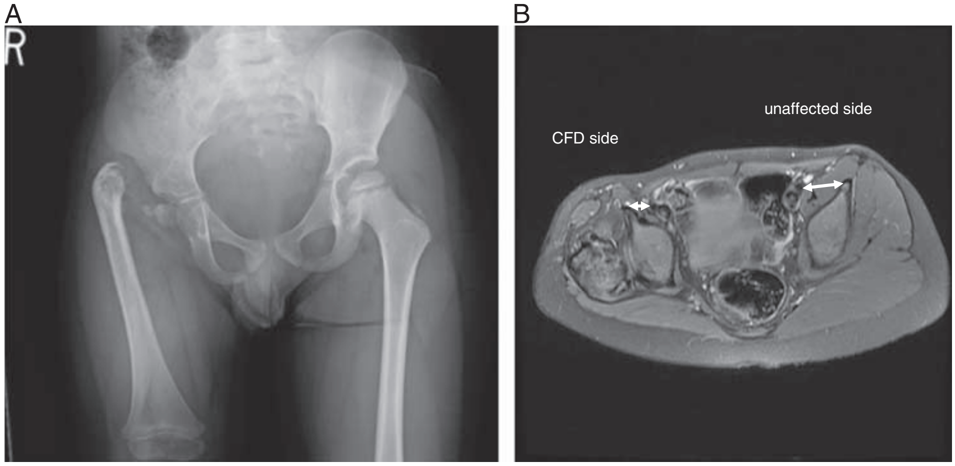

FIGURE 2. Radiologic images of a boy (age, 5 y and 5 mo) with Paley 1B1 right congenital femoral deficiency. A, Plain radiograph shows a varus deformity of the proximal femur with acetabular dysplasia, and (B) magnetic resonance imaging scans show demonstrating the distance of the femoral neurovascular bundle from the anterior inferior iliac spine on the CFD side (13 mm) and the unaffected side (28 mm). CFD indicates congenital femoral deficiency.

femoral nerve before performing a tenotomy of the rectus femoris tendon. Our MRI analysis found an important relationship between the NV bundle and AIIS of the affected side (13 mm) that was significantly different from the unaffected side (23 mm). The data analyzed did not contain any examples of the NV bundle crossing the AIIS; however, the data showed that the bundle is closer to the AIIS on the involved side when compared with the contralateral side. Because of this relationship, we recommend identifying the femoral nerve before transection of the rectus femoris tendon to ensure a safe tenotomy (Fig. 2).

The Paley classification was utilized in our study because it is based on pathologic factors and guides the surgeon in reconstructive strategies.[12] Previous classifications do not have a detailed understanding of the underlying anatomic abnormalities as the Paley classification.[1,13] All patients included in our study were classified as either Paley type 1 or 2. As discussed above, the MRI was used to give us detailed information on the femoral neck morphology. The Paley classification has a weak, inverse correlation with the AIIS percent change and the neck-shaft angle. As the classification increases in number, the femoral NV bundle moves closer to the AIIS (relative to the contralateral bundle) and the neck-shaft angle decreases (increasing varus). However, this study only evaluated Paley types 1 and 2 and their subtypes; this information should not be extrapolated onto Paley types 3 and 4.

Subsequently, we discovered both a positive correlation between the AIIS absolute distance ($R = 0.458$) and the AIIS percent change ($R = 0.798$) with the neck-shaft angle. The proximal femoral deformity seen in the coronal view on radiographs or MRI is a combination of femoral varus and soft tissue abduction/flexion contractures. We chose to use the neck-shaft angle as it might obviate the need to obtain a preoperative MRI and alert the surgeon of the abnormal location of the NV bundle location. In this patient series, as the neck-angle decreases (worsening varus) so does the distance between the femoral NV bundle to the AIIS. In addition, this relationship becomes stronger when we look at the AIIS percent change. The mean AIIS percent change was calculated to be −40%, meaning the NV bundle was 40% closer to the AIIS when compared with the contralateral side. Surgeons performing reconstructions of the hip and proximal femur in patients with CFD should be aware of this relationship. We hypothesize the forces that create the varus deformity of the proximal femur may also be responsible for the lateral migration of the femoral NV bundle toward the AIIS. Although our study does not prove a causal relationship between the neck-shaft angle and the absolute distance, there seems to be some causation between the neck-shaft angle measured on coronal plane imaging and the percentage change relative to the contralateral side ($R^2 = 0.64$).

Our study was not without limitations. We evaluated 33 patients with a relatively rare disease, but we excluded a large number of patients because of the paucity of MRI studies. At our institution, we receive many patients from throughout the world, and they often bring MRI scans from different institutions, which leads to variabilities in MRI protocol. The position of the hips was a natural, resting position. Patients with CFD have deformities (soft tissue flexion, abduction, external rotation, and boney extension, adduction, external rotation) that affect the ipsilateral hip but do not affect the unaffected side. Therefore, the affected and unaffected hips were not in the same position during the study. In addition, the MRI studies did not include contrast. In 2009, Chomiak et al[14] reported on 2 patients with CFD and variations in vascular patterns who showed internal iliac artery branches supplying the lower leg except the anterior part of the thigh and pseudarthrosis. We found we were able to identify the femoral NV bundle without contrast. However, the addition of contrast may allow for a detailed evaluation of anatomic variation of the vessels and improvement of measurement accuracy.[14]

In conclusion, our study was the first investigation that found the distance of the femoral NV bundle to surgically important landmarks was different on the affected side when compared with the unaffected side in patients with CFD. MRI of the pelvis may be considered before reconstruction to assess the location of the femoral NV bundle relative to the AIIS; however, worsening varus deformity on coronal plane imaging may also alert the surgeon to this anomaly. We recommend identifying and protecting the femoral nerve before sectioning the proximal rectus femoris tendon to ensure a safe surgical reconstruction.

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