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Evaluation of Femoral Anteversion and Femoral Neck-Shaft Angles in Cerebral Palsy and a Review of the Literature

Serebral Palsili Pediatrik Hastalarda Femoral Anteversiyon ve Femur Boyun-Şaft Açılarının Değerlendirilmesi ve Literatürün Tekrar Değerlendirilmesi

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ABSTRACT

Aim: Cerebral palsy (CP) is a chronic, sensorimotor disease as a result of damage in the brain that has not completed its development. In this study, we evaluated femoral anteversion (FA) and femoral neck-shaft (FNS) angles in pediatric patients with CP and studied whether there was a difference between the healthy population and patient group according to involvement subtypes or Gross Motor Functional Classification System (GMFCS).

Materials and Methods: Thirty patients (20 females, 10 males; mean age 10 years 8 months; range 4 to 14 years) diagnosed with spastic CP, who had undergone surgery, were evaluated retrospectively. Of thirty patients, eleven were quadriplegic, ten were diplegic and nine were hemiplegic. Fifty-one hips of 30 patients were investigated in the study. According to the GMFCS, there were nine patients at level II, eight at level III, six at level IV, and seven at level V. X-rays and computed tomograhy images of the patient and control group were evaluated by radiological files of the hospital. FA and FNS angles of all patients were measured and the obtained values were compared.

Results: The FNS angle ($p<0.05$) and the FA angle ($p<0.05$) were significantly larger in the patient group than in the control group. The FNS angle ($p<0.05$) and the FA angle ($p<0.05$) were significantly larger in the quadriplegic group than in the diplegic and hemiplegic groups. The FNS angle ($p<0.05$) and FA angle ($p<0.05$) were significantly larger in GMFCS level IV/V patients than in GMFCS level II/III patients.

Conclusion: FA and FNS angles are increased in CP compared to normal population due to the spasticity in some muscles acting on the hip and the resulting imbalance in muscle strength. In quadriplegics, angles are significantly higher compared to diplegics and hemiplegics, GMFCS level IV/V had higher angle values compared to GMFCS II/III.

Keywords: Cerebral palsy, femoral anteversion angle, femoral neck-shaft angle

ÖZ

Amaç: Serebral palsi (SP), gelişimini tamamlamamış beyin hasarı sonucu oluşan kronik, sensorimotor bir hastalıktır. Bu çalışmada pediatrik SP'li hastalarda femoral anteversiyon (FA) ve femur boyun-şaft (FNS) açılarını değerlendirdik ve sağlıklı popülasyon ve hasta grubunun kendi içinde tutulum alt tiplerine ve Kaba Motor Fonksiyonel Sınıflandırma Sistemi'ne (GMFCS) göre bir fark olup olmadığını araştırdık.

Gereç ve Yöntem: Spastik SP tanısı alan ve cerrahi uygulanan 30 hasta (20 kadın, 10 erkek; ortalama yaş 10 yıl 8 ay; dağılım 4-14 yıl) geriye dönük olarak değerlendirildi. Çalışmada 30 hastanın 51 kalçası incelendi. Otuz hastanın 11'i kuadriplejik, 10'u diplejik ve dokuzu hemiplejikti. GMFCS'ye göre II. düzeyde dokuz, III. düzeyde sekiz, IV. düzeyde altı ve düzey V'te yedi hasta bulunmaktaydı. Hasta ve kontrol grubunun bilgisayarlı tomografi görüntüleri hastanenin radyolojik dosyalarından değerlendirildi. Tüm hastaların FA ve FNS açıları ölçüldü ve elde edilen değerler karşılaştırıldı.

Bulgular: FNS açısı ($p<0,05$) ve FA açısı ($p<0,05$) hasta grubunda kontrol grubuna göre anlamlı olarak daha büyüktü. Kuadriplejik hastalarda FNS açısı ($p<0,05$) ve FA açısı ($p<0,05$) diplejik ve hemiplejik gruba göre anlamlı olarak daha büyüktü. FNS açısı ($p<0,05$) ve FA açısı ($p<0,05$), GMFCS seviye IV/V hastalarında, GMFCS seviye II/III hastalarına göre anlamlı olarak daha büyüktü.

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Sonuç: Kalçaya etki eden bazı kaslarda spastisite ve buna bağlı olarak kas gücünde dengesizlik nedeniyle SP'de normal popülasyona göre FA ve FNS açıları artar. Kuadriplejiklerde açılar diplejiklere ve hemiplejiklere göre belirgin olarak daha yüksektir, GMFCS seviye IV/V, GMFCS II/III'e göre daha yüksek açı değerlerine sahiptir.

Anahtar Kelimeler: Serebral palsi, femoral anteversiyon açısı, femur boyun-şaft açısı

INTRODUCTION

Cerebral palsy (CP) is proposed as a group of permanent disorders of the development of movement and posture, causing activity limitation, whereas the disturbances in fetal or infant brain are non-progressive. In CP, the spinal cord and muscles are structurally and biochemically normal so called all of the pathology is attributed to brain. The abnormality of the brain results in motor impairment[1].

The prevalence of CP is 2 in 1000 live births and the most common type of involvement is spastic diplegia constituting 80% of the patients[2]. The lesion in the brain creates problems in muscle tone and coordination. Over time, secondary disorders in the form of joint contractures develop due to unbalanced distribution of muscle strength in the musculoskeletal system[1-3].

Spasticity, which is an increase in the physiological tension of the muscle to passive movements, is caused by lesions in the cerebral cortex. Although the lesion in the brain does not show progression, adaptive changes secondary to spasticity occur over time in the musculoskeletal system[4].

Although muscle spasticity and contractures need physical therapy or surgical treatments like tendon lenghtenings and contracture releases for treatment, femoral anteversion (FA) and femur neck-shaft angle (FNS) increment of which are commonly seen in CP patients, leading to joint luxations or gait imbalance need purely surgical corrections for treatment[1,5]. Being aware of these angular changes, which can be detected by various measurement methods and corrected surgically, is extremely important in terms of the course of disease and determining the degree of planned surgical corrections to prevent hip instability and as well as hip joint luxation[4].

The Gross Motor Functional Classification System (GMFCS) is the most reliable classification system of CP patients according to capacity of mobility and independent movement and have been described for various bands[6].

The aim of this retrospective study is to evaluate FA and FNS angles of CP patients by comparing a healthy control group and also comparing the same angles within the patient group according to GMFCS levels.

MATERIALS AND METHODS

Files of sixty patients (32 females, 28 males; mean age 11 years 2 months; range 4 to 22 years) with a diagnosis of spastic CP who had already been operated for surgical correction of FA and FNS angles between February 2000 and July 2004 were retrospectively evaluated after the approval of the Tekirdağ Namık Kemal University of Ethics Committee (protocol number: 2021.115.04.10, date: 27.04.2021).

In order to make the evaluation as objective as possible, patients who had no visual and cooperative problems, no significant mental retardation, and no extrapyramidal system involvement were chosen. Furthermore, patients using medications affecting motor functions (e.g. baclofen, botulinum toxin, and benzodiazepine) were not included in the study. At the end, thirty patients (20 females, 10 males; mean age 10 years 8 months; range 4 to 14 years), who met the criteria and had both X-rays and computed tomography (CT) images that had already been taken as a matter of fact before their surgeries to plan the correction amount, were included in the study. A control group was formed with 30 age-matched patients (18 females, 12 males; mean age 10 years 3 months; range 4 to 16 years) having no history of CP and being admitted to the emergency department of the hospital because of polytrauma and having undergone AP pelvic X-ray and whole body CT during evaluation. Their pelvic X-rays and CT views were investigated through radiological files of the hospital.

According to involvement subtypes, eleven were quadriplegic, ten were diplegic and nine were hemiplegic. While both hips of diplegic and quadriplegic patients were included in the study, only the involved hips of the hemiplegic patients were added. As a sum, 51 hips of 30 patients were investigated in the study.

According to the GMFCS, there were nine patients at level II, eight at level III, six atlevel IV, and seven at level V.

Twenty two of the patients in the study group were born before 36th gestational week, 4 had prenatal complications, and the other 4 patients had complications in 3 months and further after birth. So called prematurity was the most seen etiologic factor in the patient group.

On the AP pelvic X-rays, a straight line was drawn from the midline of the femoral neck by accepting the midline according to two reference points. The femoral anatomical axis was also drawn and the angle at the medial side of the two lines was measured with a goniometer, providing the FNS angle (Figure 1)[7].

In order to measure the FA angle, the longitudinal axis of the femoral neck was drawn in the scan where the femoral neck appeared longest in the hip (Figure 2) in CT scans. Then, the bicondylar axis was drawn in the femoral condylar region (Figure 3). The angle of the bicondylar axis with the longitudinal axis of the femoral neck was measured[8]. This angle provided the FA value.

Data related to the study and control groups are given in Table 1 and Table 2, respectively.

For all comparisons, only the involved sides of hemiplegic patients were included in the study group (11 quadriplegic patients-22 hips, 10 diplegic patients-20 hips, 9 hemiplegic patients-9 hips; a total of 30 patients-51 hips).

Patients in the study and control groups were compared using the parameters listed below.

1. Homogeneity of variables in the study and control groups,

2. Age distribution between the study and control groups,

3. FNS and FA angle values between the study and control groups,

4. Comparison of FNS and FA angle values in the study group based on CP involvement types,

5. Comparison of FNS and FA angle values in the study group based on GMFCS types.

Statistical Analysis

To evaluate the number of the patients in the study group, G*power programme was used. The sample size was evaluated by large effect size (d=0.8) and the mean error level of α=0.05 and at the end of analysis, it was evident that at least 52 people had to be involved in the study, 26 for the study group and another 26 for the control group. For this reason, the sample size of the research was determined to be at least 52. Within the scope of data analysis, independent samples t-test, which is one of the parametric tests, was used while comparing the experimental and control groups according to variables. Before analyzing, Q-Q plot and P-P plots were examined for the assumption of normality, and it was seen that the normality assumption was met. For the assumption of homogeneity of variances, the Levene's test value was examined and it was found that the variances were not homogeneous.

For this reason, the results of the Wald test were used to calculate the t value.

The analyses were carried out in the PASW statistics 18 (2009; SPSS Inc. Chicago, IL, ABD) program and the level of significance was determined as α=0.05.

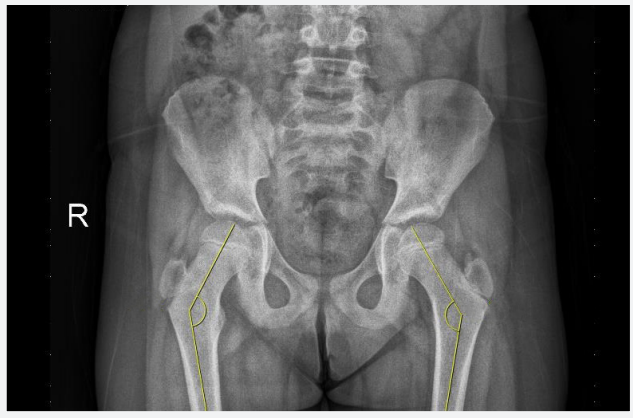

Figure 1. Femoral neck-shaft angle measurement in pelvis AP X-ray in a qudriplegic patient. Right side is measured as 143 degree, left side is measured as 141 degree

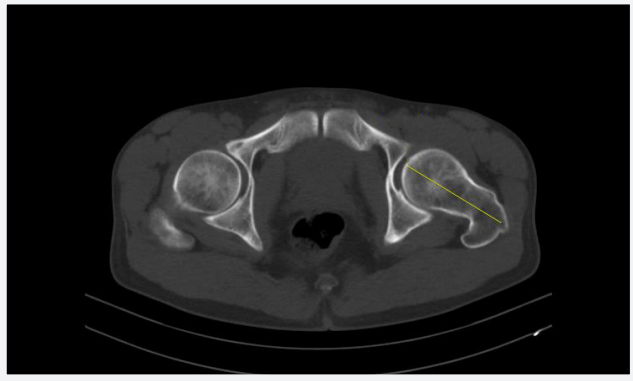

Figure 2. Reference points for left hip in computed tomography scan for measuring femoral anteversion

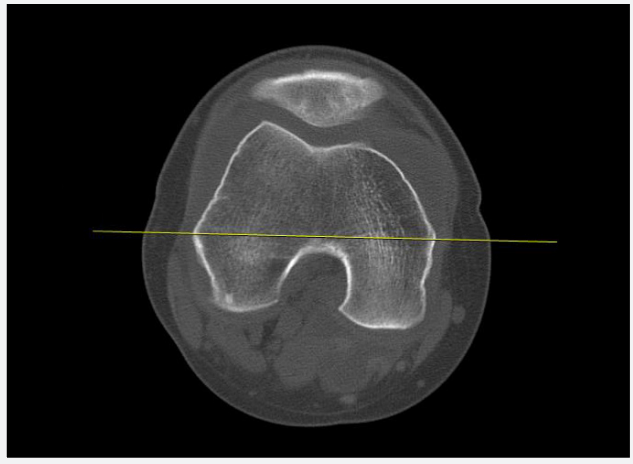

Figure 3. Reference points for knee in computed tomography scan for measuring femoral anteversion

RESULTS

All variables in the study and control groups were found to be homogeneous ($p<0.05$). There was no significant difference between the study and control groups in terms of age distribution ($p>0.05$). The FNS angle ($p<0.05$) and the FA angle ($p<0.05$) were significantly larger in the patient group than in the control group. The FNS angle ($p<0.05$) and the FA angle ($p<0.05$) were significantly larger in the quadriplegic group than in the diplegic and hemiplegic groups. The FNS angle ($p<0.05$) and FA angle ($p<0.05$) were significantly larger in the GMFCS level IV/V patients than in the GMFCS level II/III patients.

All of the variables and standard deviations are seen in Table 3.

DISCUSSION

Adduction, flexion and internal rotation deformities are the most commonly seen deformities in the hips of CP patients[1]. The type of deformity in CP patients varies depending on the type of involvement as well as the severity and distribution of spasticity. Since spasticity is more severe in quadriplegic and diplegic patients compared to hemiplegic patients, deformities may also be more severe in these patients[9]. In our study, we also revealed that quadriplegic ones had more bony deformities than the others in the patient group.

At birth, every infant, even CP patients, have a FA angle of about 40 degrees[10]. During the development process, in a healthy child, FA and its angle gradually decrease in accordance

Table 1. Patient group data

	Age	Type	Femoral anteversion angle (degrees)		Femoral neck-shaft angle (degrees)		GMFCS level
			Right	Left	Right	Left	
1	4	Quadriplegic	24	28	150	154	V
2	5	Right hemiplegic	29	15	146	138	II
3	5	Quadriplegic	37.6	38.8	142	142	IV
4	6	Quadriplegic	56	44	152	156	IV
5	6	Diplegic	50	50	142	142	III
6	7	Quadriplegic	30	34	158	162	V
7	8	Left hemiplegic	22	31	136	142	V
8	9	Quadriplegic	30	26	152	150	III
9	9	Diplegic	26	34	142	142	III
10	9	Right hemiplegic	21	10	142	136	II
11	9	Diplegic	32	30	140	142	II
12	9	Right hemiplegic	33	21	142	136	IV
13	10	Diplegic	45	41	142	142	IV
14	10	Quadriplegic	21 5	29 5	160	158	III
15	10	Diplegic	64	53	140	140	V
16	11	Diplegic	32	25	140	140	III
17	11	Right hemiplegic	40	26	142	128	II
18	11	Quadriplegic	40	50	140	140	V
19	12	Diplegic	49	46	141	142	II
20	12	Left hemiplegic	19	37	135	142	III
21	12	Quadriplegic	21	38	142	141	IV
22	12	Left hemiplegic	18	42	136	142	II
23	13	Diplegic	31	28	143	141	II
24	13	Diplegic	39.5	42.5	138	138	III
25	13	Quadriplegic	32	30	140	140	IV
26	13	Left hemiplegic	14	20	132	142	II
27	14	Quadriplegic	40	46	156	161	V
28	14	Left hemiplegic	14	26	132	142	II
29	14	Quadriplegic	30	32	139	138	V
30	14	Diplegic	53.5	52	138	140	III

GMFCS: Gross Motor Functional Classification System

with the Wolf's law due to the extensive tension of the muscle forces to the proximal femur and the plasticity of the skeletal system in the child until the age of 16 years, and decrease to the physiological level of 15 degrees in adults[10]. Whereas in CP patients, FA angle remains the same and high as a value. The main reason for the increase in FA angle in patients with CP is increased spasticity in the iliopsoas muscle[7]. Another responsible factor is the partially or completely diminished strength of the gluteal muscles adhering to the apophysis of the trochanter major[11-14]. In our study, we also concluded that FA angle was higher in the patient group compared to the healthy control group.

Although FNS angles of healthy children and children with CP are similar in the first years of life, the angles of children with CP barely change or even increase while the angles of healthy children decrease as they progress towards adult age[10]. Miller et al.[9] declared that increase of FNS angle was associated with the shortening of the femoral neck due to an increase in FA. We also declared that FNS angles were higher in the study group compared to the control group.

FA and FA angle can be measured with a numerous methods including physical examination[5,15,16], biplanar radiology X-ray[17,18], fluoroscopy[10], ultrasonography[9], 2 or 3-dimensional CT[15,19-21], and magnetic resonance imaging[22].

There are some methodological interactions for the evaluation or validation of FA angle. Chung et al.[23] revealed that physical examination combined with AP pelvic X-ray was enough for FA angle evaluation and this diminished the need for CT and excess radiation.

Table 2. Control group data

	Age	Femoral anteversion angle (degrees)		Femoral neck-shaft angle (degrees)	
		Right	Left	Right	Left
1	4	30	30	132	132
2	4	30	30	130	130
3	6	27	27	132	132
4	6	27	27	134	134
5	7	24	24	132	132
6	7	23	23	130	130
7	7	24	24	132	132
8	7	23	23	128	130
9	8	24	24	130	130
10	8	25	25	130	130
11	8	24	24	130	130
12	9	21	20	130	130
13	9	22	22	128	130
14	9	22	22	132	130
15	9	21	21	130	130
16	9	22	22	128	128
17	10	21	21	128	128
18	10	21	20	130	128
19	11	20	21	129	129
20	11	20	21	130	130
21	12	20	20	129	129
22	12	20	20	130	130
23	12	20	20	129	129
24	12	21	21	130	130
25	13	20	21	130	130
26	13	20	20	129	129
27	14	15	16	130	130
28	14	15	16	129	129
29	16	14	14	129	129
30	16	15	15	130	130

Table 3. Femoral anteversion angles and femoral neck-shaft angles of patient group according to cerebral palsy type				
	N (patients)	N (hips)	Femoral anteversion angle (degrees)	Femoral neck-shaft angle (degrees)
Total	30	51	58.50±33.67 (15.50-58.50)	142.98±6.91 (135-160)
CP type				
Quadriplegic	11	22	34.47±8.29 (25.50-50)	148.77±8.52 (138.50-160)
Hemiplegic	9	9	24.33±6.01 (15.50-33)	138.39±1.93 (135-142)
Diplegic	10	20	41.18±10.96 (28.50-58.50)	140.75±1.44 (138-142)
GMFCS				
II/III	17	27	31.13±10.92 (15.50-52.75)	141.35±5.71 (135-159)
IV/V	13	24	36.98±10.22 (26-58.50)	145.12±7.94 (138.50-160)
GMFCS: Gross Motor Functional Classification System, CP: Cerebral palsy				

Miller et al.[24] revealed that ultrasonography of the hip performed in internal rotation position gave better results than CT. In an experimental investigation on femora models, Riccio et al.[25] found that 3D reconstruction of CT was necessary and gave more accurate results about FA angles independent of FNS angles.

Davids et al.[21] revealed that the most important was the position of the patient during CT scanning. If the position is inaccurate then even 3D reconstruction will give false results vice versa 2D CT is enough for correct results.

In case of FNS angles, Kay et al.[26] revealed in a cadaveric study that positioning of the extremity in 15 degrees of internal rotation would give the most accurate information about FNS angle.

Davids et al.[15] reported that trochanteric prominence angle test and 3D CT scans gave similar increasing of FA and FNS angle results in CP patients so there was no need for CT to evaluate these patients.

In contrast to all these, Scorcelletti et al.[8] have revealed that there is not a reliable evaluation method for FA and FNS angles and CT gives high doses of radiation to patients so must not be used.

There is still a debate in the literature about the accurate measurement technique of FA and FNS in CP patients and a consensus could not been achieved yet. In our study, we used 2 dimensional CT for the measurement of FA angles and plain pelvic X-rays for FNS angle measurements.

Yamaguchi et al.[14] revealed that in non-ambulatory patients, FNS and FA were found in higher values than in ambulatory patients.

Bobroff et al.[10] stated that FA angle was higher in ambulatory CP patients compared to non-ambulatory ones but in contrast, FNS was smaller as an angle value in ambulatory ones compared to non-ambulatories, and they concluded this with fluoroscopy method.

In a biplanar radiographic study, Laplaza and Root[17] concluded that there was not any difference between ambulatory and non-ambulatory CP patients in means of FA angles but they also reported that FNS angle was higher in non-ambulatory ones.

Gose et al.[27] found with 3D CT that FA angle change was controversial but FNS angle was higher in non-ambulatory ones and they also revealed that in quadriplegic and GMFCS IV-V levels, FNS and FA angles were higher compared to diplegics and GMFCS II-III respectively.

Massaad et al.[28] revealed that FA and FNS angle increased in CP patients compared to a healthy control group and they concluded this with low dose biplanar X-ray technique.

Robin et al.[5] concluded that FA angles were higher in GMFCS I-II patients than in healthy control group but were lower than GMFCS III to V group and they also concluded that FNS angles increased as GMFCS level increased.

Laplaza et al.[7] reported that GMCSF level was the main factor affecting FA and FNS angles so called when level increased the angles also increased.

In our study, we found that FA and FNS angles were both higher in non-ambulatory group of CP patients compared to ambulatory ones by means as GMFCS level increased both of the angles also increased and we also concluded that FA and FNS angles were higher in quadriplegics compared to diplegics and hemiplegics.

Study Limitations

This article has some limitations. The age variability in a large range is a limitation. Another limitation is that measurement of FNS angle on AP X-ray alone is not sufficient, these measurements cause errors especially in the hips with increased FA. Actually, this measurement on the AP X-ray gives the projected angle, while the true angle is obtained from the intersection points of the projected FNS angle in AP pelvic and lateral hip X-rays taken at 90 degrees of hip flexion and 20 degrees of hip abduction on corrected tables[29].

CONCLUSION

CP patients are coming in front of orthopedic surgeons with increasing in number by the development and wellness of newborn intensive care units so called they care premature newborns much better compared to past. As the number of CP patients increases, their evaluation and surgeries also increase both in number and in importance.

FA and FNS angles are important for well-being of the hips of CP patients. FA and FNS angles increase in this patient group and this will bring their hips in danger for instability and luxation. If there is any suspicion of a change in FA and FNS angles in the clinical examinations performed during the periodic visits of patients with CP, these angles should be examined. If FA increase causing apparent toeing-in gait pattern in patients or FA and FNS increase showing tendency of subluxation is detected, soft tissue and osseous surgical interventions should not be avoided in order to prevent further hip pathologies. The progression of coxa valga is more critical and grave in quadriplegics and GMFCS IV/V group.

Ethics

Ethics Committee Approval: The study were approved by the Tekirdağ Namık Kemal University of Local Ethics Committee (protocol number: 2021.115.04.10, date: 27.04.2021).

Informed Consent: Retrospective study.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: G.Z., Concept: M.A., G.Z., Design: M.A., Data Collection or Processing: M.A., Analysis or Interpretation: M.A., Literature Search: M.A., Writing: M.A.

Conflict of Interest: No conflict of interest was declared by the authors.

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