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Higher femoral head–to–femoral neck ratio are associated with limited range of motion of hip joints in hereditary multiple exostosis patients

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Abstract

Hip involvement is one of the most common skeletal manifestations of hereditary multiple exostoses (HME) in 30-90% of the patients. The anatomical changes in the proximal femur in patients with HME, may lead to deformities and related functional deficits. Its effect on the range of motion (ROM) of the hip joint is not well described. The aim of this study was to evaluate the role of hip deformity of HME disease in the ROM of the hip joint. It was hypothesized that certain morphological parameters affect the ROM of the hip joint. Conclusively, 44 patients including 27 men and 17 women (88 hips) were evaluated. Hips are divided in Group 1 (no ROM limitation group) and Group 2 (ROM limitation group). The head-neck ratio (HNR), center edge angle (CE), Sharp's acetabular angle (SAA), alpha angle (AA), neck shaft angle (NSA) and ROM parameters were measured. The correlations between radiological parameters (SAA, CE, NSA, AA and HNR), and the ROM data (F, E, Abd, Add, IR and ER) were statistically evaluated. All evaluated parameters (SAA, CE, NSA, AA) except the HNR do not show significant differences between study group 1 (no ROM limitation group) and group 2 (ROM limitation group). The means of the HNR in group 1 were significantly higher than those in group 2 (HNR: 1.23 vs. 0.92, $p<0.001$). In group 2 HNR showed a strong positive correlation with Abd (PC 0.72, $p<0.001$), E (PC 0.7, $p<0.001$), ER (PC 0.67, $p<0.001$), IR (PC 0.67, $p<0.001$) and F (PC 0.66, $p<0.001$). HME patients with hip involvement have better ROM of the hip joint, if they have the higher HNR. Additionally, a positive correlation between HNR and ROM capacity was determined.

Keywords: Hereditary multiple exostoses, femoral head-neck ratio, acetabular angle, center edge angle, femoral neck-shaft angle, alpha angle

Introduction

Hereditary multiple exostosis (HME) is characterized by multiple osteochondromateous proliferation within the perichondral ring of the growth plates of long bones, ribs, hip, and vertebrae [1]. Regarding the considerable amount and localization, osteochondromas may cause accompanying conditions [1-4]. Hip involvement is one of the most common skeletal manifestations of HME in 30-90% of the patients [5].

It is stated that the anatomical changes in proximal femur in patients with HME, may lead to deformities and related functional deficit [6,7]. The femoral flaring, as assessed by the femoral head–to–femoral neck ratio (HNR), Sharp's acetabular angle (SAA), the center edge angle (CE), the femoral neck-shaft angle (NSA), the alpha angle (AA) have been introduced as morphological parameters that can evaluate severity of the HME patients' hip deformity. However, there are limited number of studies evaluating the relationship between the degree of the hip deformity and the range of motion (ROM) of the hip joint in this specific condition [8,9].

The aim of this study was to evaluate the role of hip deformity of HME disease in the ROM of hip. It was hypothesized that certain morphological parameters effect the ROM of HME patients' hip.

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Material and Methods

After obtaining institutional review board approval in Health Science University Baltalimanı Bone Diseases Education and Research Hospital (approval ID number: 17022015/20), we identified 135 patients with HME who underwent treatment between January 1995 and May 2015. A total of 91 patients with a history of a hip surgery (n=31), the limb length discrepancy (n=15), neuromuscular dysfunction (n=3), skeletally immature patients (n=39) and hip joint arthritis (n=3) patients were excluded. Conclusively, 44 patients including 27 men and 17 women (88 hips) were evaluated. The demographic data were obtained from a review of the medical records.

The measurements were performed on the Infinitt PACS (Infinitt Healthcare Co., Seoul, South Korea) digital imaging system. Angular measurements were performed on anteroposterior (AP) and axial pelvic radiographs.

To assess the ROM of the hip joints, a manual goniometer was employed. This examination was carried out by two experienced orthopedic surgeons. Flexion (F) was assessed while the patient was lying supine with the hip positioned at 0 degrees of abduction, adduction, and rotation. Extension (E) was evaluated in the prone position with the hip also at 0 degrees of abduction, adduction, and rotation [10]. Passive ROM for hip internal rotation (IR) and external rotation (ER) was assessed while the participant was lying face down on an examination couch. During this evaluation, the first examiner held the hip steady with one hand and gently rotated it passively with the other, while the second examiner recorded the measurements. To measure hip adduction (Add) and abduction (Abd), the assessment started from the neutral position, where the thigh's longitudinal axis was aligned perpendicular to a transverse line drawn across the anterior superior iliac spines of the pelvis. These anatomical landmarks were also used to align the stationary arm of the goniometer. The goniometer's fulcrum was centered using the unilateral anterior superior iliac spine, and the moving arm was positioned over the femur's midline, pointing toward the center of the patella. To ensure stability during abduction and avoid constraining leg movement during adduction, the participant's contralateral leg hung freely over the edge of the examination table, ensuring that the pelvis remained steady during the assessment.

Hips with ≥110° of flexion, ≥10° of extension, ≥30° of abduction, ≥40° of external rotation, ≥30° of internal rotation and ≥30° of adduction were included in group 1 as hips without limitation of ROM. All remaining hips were included in group 2 as hips with limitation of ROM [11].

A senior musculoskeletal radiologist conducted measurements on several radiological parameters. SAA is determined by measuring the angle formed between a line connecting the superolateral and inferomedial portions of the acetabulum and another line connecting the inferior tips of the pelvic teardrops (Figure 1); the CE was defined as the angle formed by a line that connects the center of the femoral head to the lateral edge of the acetabulum and another line that is perpendicular to the inter-teardrop line and passes through the center of the femoral head (Figure 2); the femoral NSA was described as the angle created by the anatomical axes of the femoral neck and shaft (Figure 3); the AA was established as the angle formed by a line extending from the anterior point, where the distance from the center of the head exceeds the radius of the subchondral surface of the femoral head, and another line drawn from the center of the head through the narrowest part of the femoral neck (Figure 4); the femoral HNR was described as the proportion between the widest measurement of the femoral head and neck (Figure 5) [12-17].

The Shapiro–Wilk test was employed to assess the normality of continuous variables. When the data were found to follow a normal distribution, Student's t-test was utilized to compare the means between group 1 and group 2. For non-continuous variables, the normality was evaluated using the Mann Whitney U test, and if the data exhibited a normal distribution, Student's t-test was again applied for mean comparisons between group 1 and group 2. To compare the distribution of sexes between groups, the chi-square test was employed. The statistical significance level was set at 0.05, and data analysis was conducted using the SPSS 21 program. At the end of the study, post-hoc analyse for the HNR was calculated as 100%. The correlations between radiological parameters (SAA, CE, NSA, AA and HNR) and the ROM data (F, E, Abd, Add, IR and ER) were statistically evaluated using Pearson correlation tests.

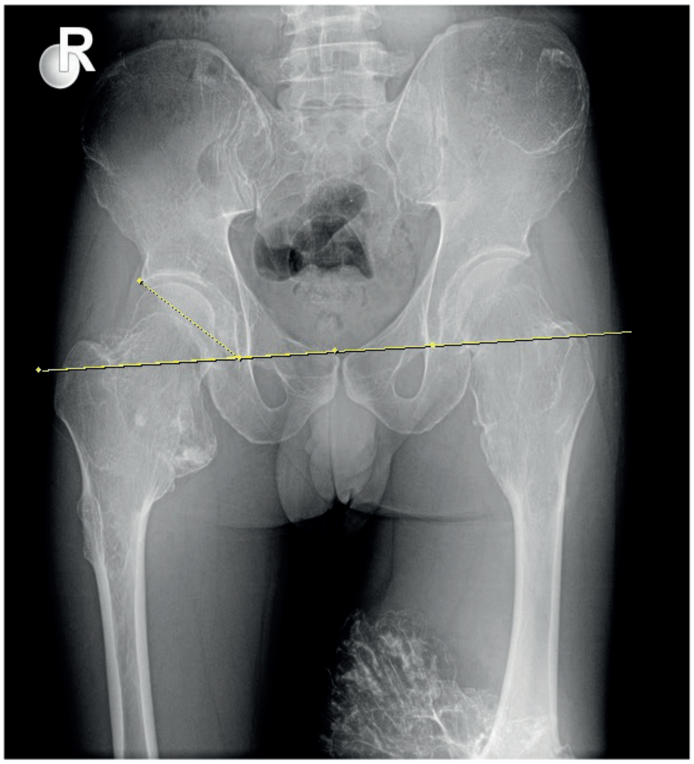

Figure 1. SAA was defined as the angle between the line joining the superolateral and inferomedial part of the acetabulum and another line joining the inferior tips of the pelvic teardrops

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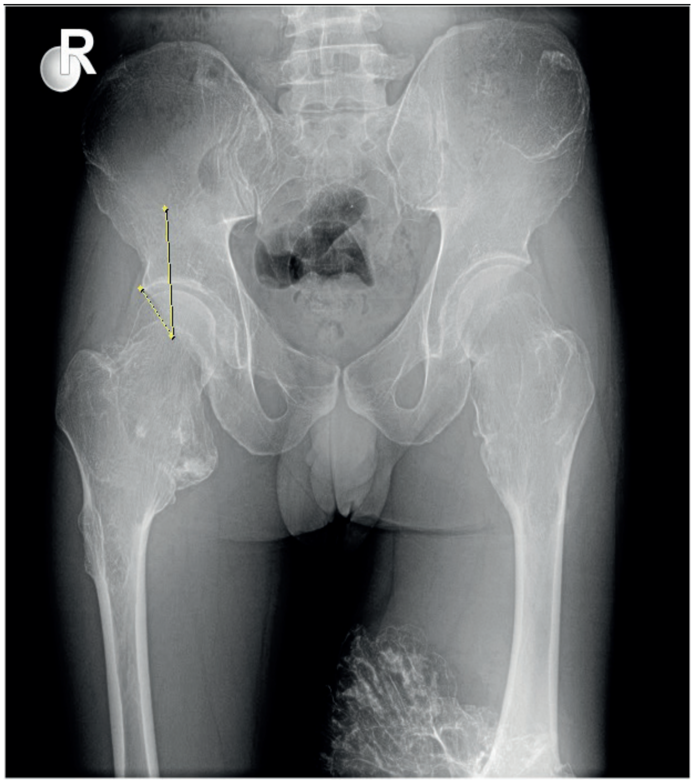

Figure 2. CE was defined as the angle between a line joining the center of the femoral head to the lateral edge of the acetabulum and a line perpendicular to the inter-teardrop line passing through the center of the femoral head

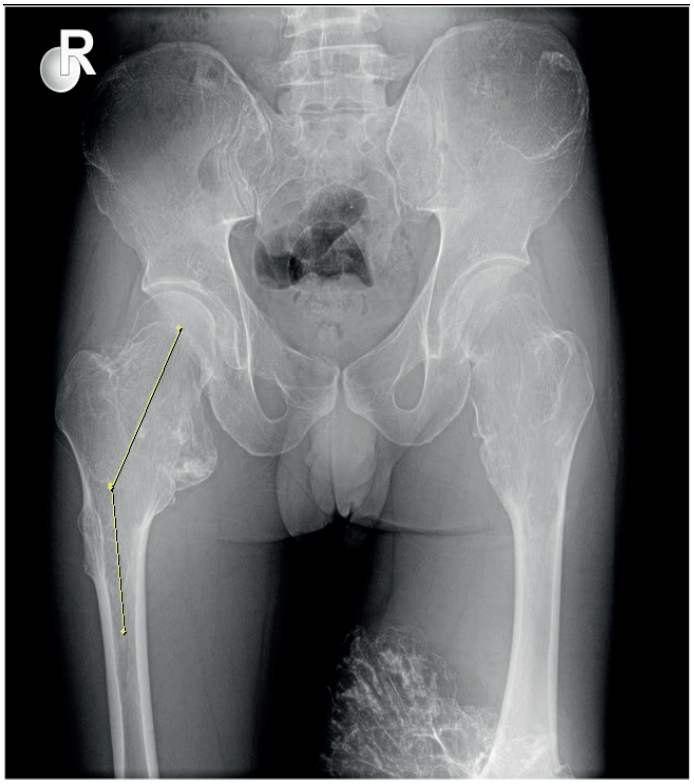

Figure 3. NSA was defined as the angle formed between the anatomic axes of the femoral neck and shaft

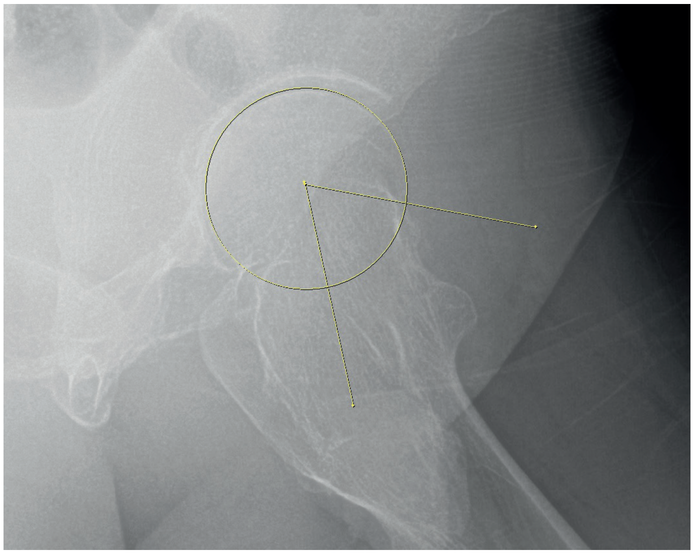

Figure 4. AA is between the line connecting the point of no sphericity of femoral head from the center of femoral head and the line extending up to the center of femoral head from the center of femoral neck

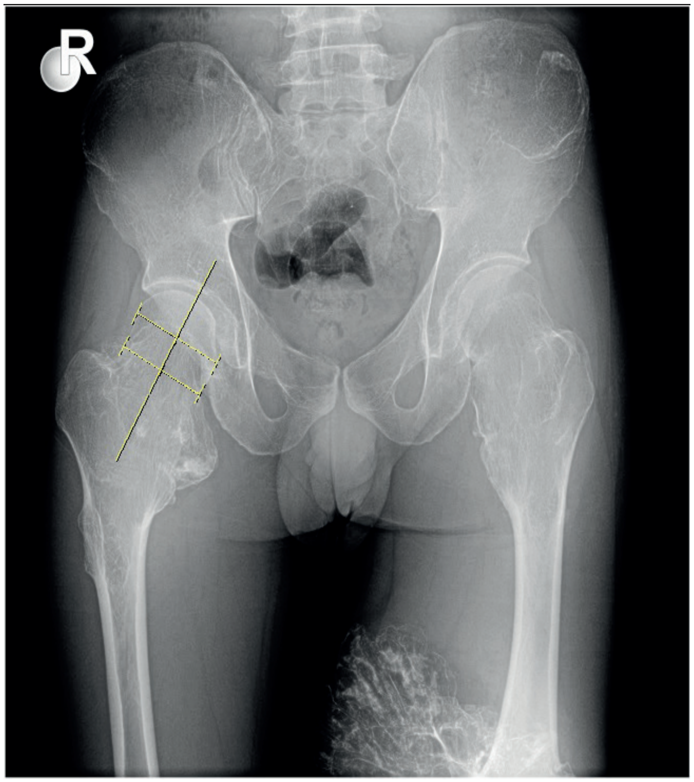

Figure 5. HNR was defined as the ratio between the widest distance of the femoral head and neck

Results

Patient characteristics were summarized in table 1. All evaluated parameters (SAA, CE, NSA, AA) except the HNR did not show significant difference between the study group 1 (no ROM limitation group) and group 2 (ROM limitation group). The results of each parameter are summarized in Table 2. The means

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HNR in group 1 were significantly higher than those in group 2 (HNR: 1.2 vs. 0.9, p<0.001). The mean SAA, CE, NSA and AA in group 1 were not significantly different than those in group 2 (SAA: 40.8° vs. 39.6°, p=0.336; CE: 35.4° vs. 36.6°, p=0.514; NSA: 142.1° vs. 144.8°, p=0,125; AA: 60.2°vs. 63.8°, p=0.057).

In group 1 HNR showed a moderate positive correlations with E (PC 0.544, p=0.007), Abd (PC 0.434, p=0.039). There is no correlation between HNR and IR, ER, Add, F. Additionally, there was no correlation between SAA, CE and the hip ROM parameters. On the other hand, there was a moderate negative correlation between NSA, AA and the hip ROM parameters (Table 3). (NSA vs. F: PC -0.468, p=0.024; NSA vs Abd: PC -0.476, p=0.022; AA vs E: PC -0.489, p=0.018; AA vs Add: PC 0.441, p=0.035).

In group 2 HNR showed a strong positive correlations with Abd (PC 0.722, p<0.001), E (PC 0.698, p<0.001), ER (PC 0.669, p<0.001), IR (PC 0.667, p<0.001) and F (PC 0.663, p<0.001). However, there was no correlation between SAA, CE and the hip ROM parameters. On the other hand, there was a poor negative correlation between NSA, AA and the hip ROM parameters (Table 4). (NSA vs. F: PC -0.272, p=0.028; NSA vs ER: PC -0.247, p=0.047; AA vs F: PC -0.269, p=0.03; AA vs Abd: -0.261, p=0.036).

Table 1. The demography of the control and patient groups

	Group 1		Group 2		
	Mean±SD	**Min-Max**	**Mean±SD**	**Min-Max**	**p**
Age	25.6±10.5	16-47	28±10.35	16-51	0.261[a]
			Gender n (%)		
Female	11 (47.8%)		23 (35.4%)		0.292[b]
Male	12 (52.2%)		42 (64.6%)		
			Side n (%)		
Right	9 (39.1%)		35 (53.8%)		<0.001[b]
Left	14 (60.9%)		30 (46.2%)		
Flexion	133.04±5.17	120-135	113.77±13.64	90-135	**<0.001[a]**
Extension	33.48±3.82	20-35	24±6.8	10-35	**<0.001[a]**
Internal rotation	36.96±3.28	30-40	26.31±10.13	0-45	**<0.001[a]**
External rotation	44.13±1.94	40-45	25±7.86	0-35	**<0.001[a]**
Abduction	43.7±3.1	35-45	30.92±9.52	15-45	**<0.001[a]**
Adduction	40.30±3.20	35-47	23.35±8	9-44	**<0.001[c]**

a: Mann Whitney U test, b: Chi-Square test, c: Student's t test

Table 2. Summary of each parameter

	Group 1	Group 2	
	Mean±SD (range)	**Mean±SD (range)**	**p-value**
SAA	40.8±5.4 (33-52)	39.6±5.3 (28-55)	0.336
CE	35.4±6.4 (18-44)	36.6±8.1 (15-52)	0.514
NSA	142.1±5.1 (131-149)	144.8±7.6 (130-166)	0.125
AA	60.2±7.2 (52-81)	63.8±8 (48-87)	0.057
HNR	1.2±0.1 (1.06-1.38)	0.9±0.2 (3.04-14.4)	<0.001

p:Student's t test, SAA: Sharp's acetabular angle, CE: center edge angle, NSA: neck shaft angle, AA: alpha angle, HNR: head neck ratio

Table 3. Correlation analysis between ROM parameters and morphological parameters of group 1

Group 1		SAA	NSA	CE	AA	HNR
F	r	0.078	**-0.468**	0.156	-0.020	0.195
	p-value	0.723	**0.024**	0.477	0.930	0.373
E	r	0.177	-0.360	0.116	**-0.489**	**0.544**
	p-value	0.419	0.092	0.597	**0.018**	**0.007**
Abd	r	0.163	**-0.476**	0.260	-0.181	**0.434**
	p-value	0.457	**0.022**	0.231	0.408	**0.039**
Add	r	-0.047	-0.025	0.030	**0.441**	-0.202
	p-value	0.831	0.911	0.891	**0.035**	0.356
IR	r	0.029	-0.053	0.267	0.206	0.125
	p-value	0.896	0.811	0.218	0.346	0.571
ER	r	0.035	-0.286	0.078	0.269	-0.095
	p-value	0.875	0.186	0.723	0.214	0.665

SAA: Sharp's acetabular angle, CE: center edge angle, NSA: neck shaft angle, AA: alpha angle, HNR: head neck ratio, F: flexion, E: extension, Abd: abduction, Add: adduction , IR: internal rotation, ER: external rotation

Table 4. Correlation analysis between ROM parameters and morphological parameters of group 2

Group 1		SAA	NSA	CE	AA	HNR
F	r	0.129	**-0.272**	-0.056	**-0.269**	**0.663**
	p-value	0.305	**0.028**	0.660	**0.03**	**0.000**
E	r	0.130	**-0.318**	-0.052	-0.211	**0.698**
	p-value	0.300	**0.010**	0.678	0.091	**0.000**
Abd	r	0.069	-0.155	-0.040	**-0.261**	**0.722**
	p-value	0.585	0.217	0.753	**0.036**	**0.000**
Add	r	0.071	0.068	-0.216	-0.135	0.000
	p-value	0.576	0.589	0.084	0.284	0.999
IR	r	0.080	-0.103	0.019	-0.239	**0.667**
	p-value	0.526	0.413	0.882	0.056	**0.000**
ER	r	0.044	**-0.247**	-0.068	-0.185	**0.669**
	p-value	0.725	**0.047**	0.590	0.141	**0.000**

SAA: Sharp's acetabular angle, CE: center edge angle, NSA: neck shaft angle, AA: alpha angle, HNR: head neck ratio, F: flexion, E: extension, Abd: abduction, Add: adduction, IR: internal rotation and ER: external rotation

Discussion

The hip involvement in HME has been reported as high as 30% of the cases [3]. However, there are few studies evaluating radiological parameters that correlate the severity of the hip ROM limitation in HME patients. In our study the relationship between SAA, CE, NSA, AA, HNR and the hip ROM limitation in HME patients were investigated.

HME typically encompass a significant portion of either the metaphysis or diaphysis and tend to have a more irregular shape [18]. Altered bone remodeling typically occurs, leading to the clubbing of the ends of long bones. This alteration results in the loss of the normal flare between the metaphysis and diaphysis, causing the femoral necks to appear shorter and broader [19]. Rarely secondary osteochondroma can be developed [20]. Stulberg et al. has first described the femoral HNR and Halle et al. has determined that decreased HNR was associated with osteoarthritis of the hip in young men [21,22]. To the best of our knowledge, this study is the only trial to investigate the relationship between HNR and ROM parameters in HME patients with hip involvement. In present study, the HNR was found significantly higher in group 1and patients with higher HNR had better ROM. The proximal femoral osteochondroplasty might be considered by hip surgeons to increase ROM of hip joint in HME patients with high metaphyseal flaring.

The normal NSA of the femur is 130°±5° [23]. The group 1 and 2 have coxa valga deformity (NSA>135°). The manifestations of hip involvement in patients with HME are reported as coxa valga, hip dysplasia and hip subluxation [7]. In a prognostic

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investigation conducted by Siebenrock et al., it was found that hips with valgus alignment and higher levels of antetorsion exhibit a primary limitation in extension, ER, and adduction compared to hips with a normal alignment. Conversely, these hips display an extended range of IR during flexion [24]. Most studies considering HME deal with just the definition the proximal femoral deformity [7,13,25]. In present study, NSA did not differ significantly between group 1 and group 2. This finding suggests that no relationship between coxa valga and ROM parameters in patients with HME can be shown.

The AA is a radiological measurement introduced for diagnosing and assessing surgical treatments in cases of cam-type femoroacetabular impingement (FAI) [26]. Cam impingement is suggested by an alpha angle greater than 50 degrees, as originally documented by Notzli et al. [17]. The upper limit of normal considered 42° by some authors [27]. The change in AA was correlated with the increase in internal rotation [28]. As far as we know, this study is the only trial to investigate the AA in HME patients with hip involvement. The mean AA values of both groups were found to be greater than 42°. There was poor correlation between Abd, Add, F and AA in group 2. Additional investigations with larger study population are needed to evaluate the influence of AA on functional status.

SAA and CE are reliable for assessing acetabular dysplasia [29,30]. Hip dysplasia is characterized by a shallow acetabulum, generally diagnosed by a lateral CE angle less than 20° and Sharp angle more than 45° using radiography [31]. In the literature it is mentioned that osteochondromas around the hip can cause acetabular dysplasia and hip joint subluxation [7,13,25]. As our study population number could be considered a large number for HME which is a rare disease and the mean CE and SAA values of both groups were found to be in normal range, further investigations with larger study population are needed to determine whether the hip involvement of the HME cause acetabular dysplasia.

This study has various limitations. To obtain a more precise assessment of whether morphological parameters contribute to limited ROM, it would be advisable to conduct a prospective longitudinal cohort study. Additionally, the study population, particularly in group 1, was relatively small.

This study determined whether previously described parameter HNR are related with ROM of hips in HME patients with hip involvement. Patients with the higher HNR have better ROM of hip joint. Assessing the HNR can be helpful for prognostic evaluation of HME patients with hip involvement.

Conclusion

This study determined whether previously described parameter HNR are related with ROM of hips in HME patients with hip involvement. Patients with the higher HNR have better ROM of hip joint. Assessing the HNR can be helpful for prognostic evaluation of HME patients with hip involvement.

Conflict of Interests
The authors declare that there is no conflict of interest in the study.

Financial Disclosure
The authors declare that they have received no financial support for the study.

Ethical Approval
After obtaining institutional review board approval in Health Science University Baltalimani Bone Diseases Education and Research Hospital (approval ID number: 17022015/20), we identified 135 patients with HME who underwent treatment between January 1995 and May 2015.

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