Radiographic Analysis of the Pediatric Hip Patients With Hereditary Multiple Exostoses (HME)

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Background: This study aimed to report the radiographic presentation of involved hips in children with hereditary multiple exostoses (HME). This included radiographic hip measurements, osteochondromas location, and relationship with hip subluxation.

Methods: Anteroposterior pelvis radiographs of children with HME, seen between 2003 and 2014, were retrospectively reviewed. Only patients who were skeletally immature at the first visit were included. One radiograph per patient per year was reviewed. Radiographs were examined for the presence of osteochondromas and their locations. Different parameters were evaluated: femoral neck-shaft angle, Reimer migration percentage (MP), Sharp acetabular angle, Wiberg angle, femoral head-neck ratio (coronal plane), and Shenton line. All measured radiographs were divided into 3 age groups: ≤ 8, > 8 and < 13, and ≥ 13 years. Differences of the measured parameters with age were evaluated. Children with hip subluxation were identified and any relationship with osteochondromas locations, as well as MP changes over time, was recorded. Radiographs of children with a minimum 2-year follow-up were identified and changes of their hip measurements and osteochondromas' presence over time were recorded.

Results: A total of 51 children (102 hips) with HME were identified. In most locations, there was an overall increase of the occurrence of osteochondromas in the older age groups. However, in the medial femoral neck, a significantly less numbers of osteochondromas were found after 13 years of age ($P = 0.018$). There was a decrease in MP with age ($P < 0.05$). There was also an increase in Sharp and Wiberg angles in the older patients ($P < 0.05$). Hips with broken Shenton line decreased in number with age (P 0.028). Hip subluxation was encountered in 23 hips. No specific location of osteochondromas was found to have a relationship with subluxation. Thirty-six children had a minimum follow-up of 2 years (mean age at first visit 8.5 y and at last visit 13.1 y). In these children, an increased occurrence of lesions was found in medial femoral neck and ischium ($P < 0.05$) between the first and the last visit.

Conclusions: In children with HME, radiographic evaluation of the hip is necessary based on the high percentage of hip involvement. When hip osteochondromas are found, radiographic surveillance is recommended to detect hip subluxation. Surgery may certainly be necessary for symptomatic osteochondromas. However, given the possibility of improvement in hip parameters with age, early surgical treatment to improve hip longevity does not seem to be warranted.

Level of Evidence: Level IV—prognostic study.

Key Words: hereditary multiple exostoses, hip, osteochondroma

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Hereditary multiple exostoses (HME) is an autosomal dominant disorder that manifests as multiple benign tumors (osteochondromas) in the metaphyses of long bones.[1,2] The disorder has a prevalence of 1 per 50,000 in the general population[2–4] and has a variable degree of expression between individual members of the same family.[1] The lesions usually are not present at birth, but appear between the ages of 2 and 10 years.[1,4,5] They increase in size during childhood, and stop growing at skeletal maturity; after which no new lesions develop.[5,6] However, the possibility of spontaneous resolution has been described with both solitary as well as multiple osteochondromas.[4,7–10]

In addition to long bones, other locations that may be affected include the ribs, spine, scapula, and pelvis; sometimes in a symmetric pattern.[5] Hip osteochondromas are present in a high percentage of HME cases, mainly in the proximal femur, where they have been reported in 30% to 90% of patients.[3,11,12] Although osteochondromas around the hip are often asymptomatic,[13] they can produce abnormal mechanical forces which may lead to hip dysplasia, subluxation, impingement, labral tears, and premature osteoarthritis.[12,14,15]

The evolution of hip deformities, seen in children with HME, is still not clear. Ozaki et al[16] found that the femoral neck-shaft angle tended to decrease with age, making the surgical treatment unnecessary during growth. Conversely, other studies have suggested that these hip deformities tend to worsen with age[13] and early surgical treatment was recommended.[11,13,17–20]

The aim of this study was to report the radiographic presentation of involved hips in children with HME.

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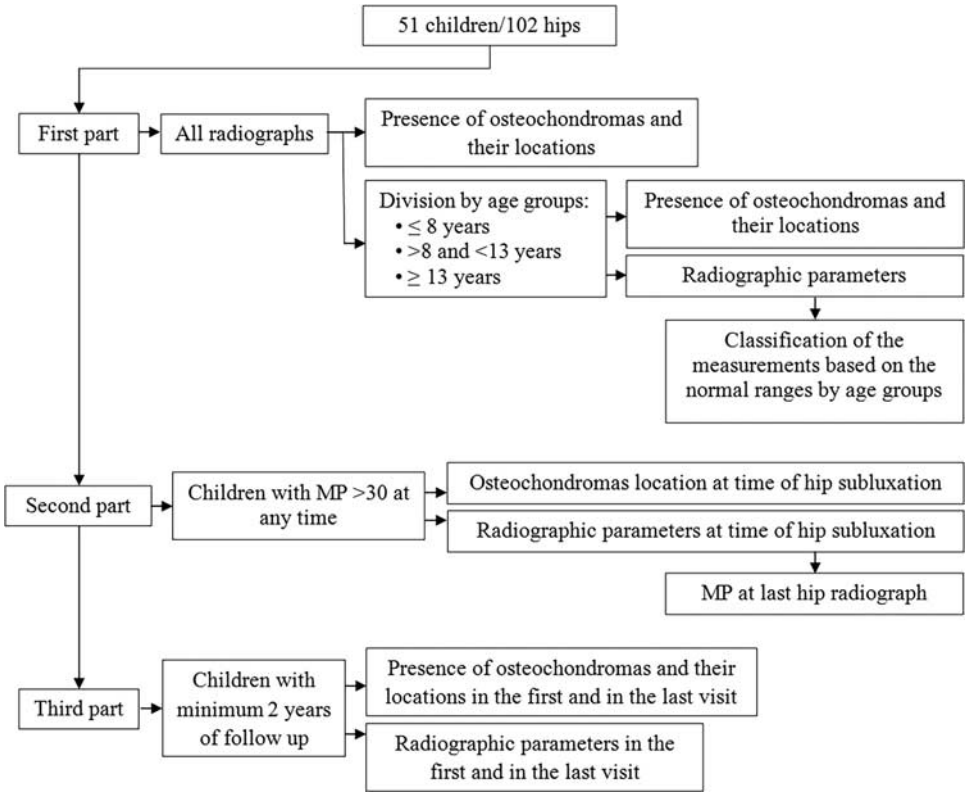

FIGURE 1. The study design showing the parameters evaluated in each part of the study. MP indicates migration percentage.

This included radiographic hip measurements, osteochondromas locations, and relationship with hip subluxation.

METHODS

After obtaining the approval of our Institutional Review Board; records of all patients with HME seen between 2003 and 2014, were retrospectively reviewed. Demographic data were collected. All patients who were skeletally immature (growth plates were visible on radiographs), with at least 1 anteroposterior (AP) pelvis radiographs, were included. In the cases where the same patient had more than 1 AP pelvis radiograph; only 1 radiograph per year was reviewed, starting with the first available radiograph. In the surgical cases, only the preoperative radiographs were included. All images were evaluated by 1 orthopaedic surgeon and different measurements were used to give a comprehensive description of the hip morphology.

The analysis was planned in 3 parts (Fig. 1); the first part was a cross-sectional analysis that included all measured radiographs. Radiographs were examined for the presence of osteochondromas and their locations. Subsequently, the radiographs were divided into 3 age groups: ≤ 8, >8 and <13, and ≥ 13 years. The age groups were selected so they have a similar number of radiographs. The presence of osteochondromas was recorded in each age group to compare differences with age. Radiographic measurements included the femoral neck-shaft angle,[15,21–23] Reimer migration percentage (MP),[15,24] Sharp acetabular angle,[15,25] Wiberg center-edge angle,[14,15,26] and femoral head-neck ratio (coronal plane).[15] Disruption of Shenton line was recorded as well. Comparisons were performed between the age groups.

Within each age group, hips with measurements in the normal range were identified. Normal ranges are as follows: femoral neck-shaft angle: ≤ 135 degrees for children ≥ 12 years and ≤ 150 degrees for younger children[15,21,22]; Sharp acetabular angle: ≤ 45 degrees[15]; Wiberg angle: ≥ 20 degrees[15,26]; and Reimer migration: normal: $<20\%$; borderline: 20% to 29%, subluxation: $>30\%$.[15,24]

In the second part of the study (Fig. 1), children with hip subluxation, at any time during the study period, were identified according to their MP ($>30\%$).[3,15] Osteochondroma locations and radiographic measurements were recorded at the time of hip subluxation diagnosis for each patient. MP on the last available radiograph was recorded as well.

The third part of the analysis (Fig. 1) included longitudinal data from children with a minimum 2-year radiographic follow-up. Radiographic measurements, as well as any change of osteochondromas' presence over time, were recorded. In this subgroup of patients, a comparison between the first and the final radiograph was made.

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TABLE 1. Comparison of the Osteochondromas Presence at Different Locations Between Age Groups

	N (%)			Comparison Between Age Groups (*P*)			
Location	**≤8 y (102 Hips)**	**> 8 to < 13 y (102 Hips)**	**≥ 13 y (120 Hips)**	**≤8 vs. > 8 to < 13 y**	**≤8 vs. ≥ 13 y**	**> 8 to < 13 vs. ≥ 13 y**
Medial femoral neck	59 (58)	78 (76)	74 (62)	0.005*	0.562	0.018*
Lateral femoral neck	54 (53)	62 (61)	63 (53)	0.258	0.948	0.215
Ischium	37 (36)	53 (52)	62 (60)	0.024*	0.0004*	0.229
Pubis	15 (15)	27 (26)	46 (38)	0.038*	0.0001*	0.061
Acetabulum	11 (11)	25 (25)	17 (14)	0.010*	0.449	0.050*
Femoral head	9 (9)	6 (6)	4 (3)	0.421	0.083	0.361

χ^2 test was used. Level of significance was set as 0.05.
*P value <0.05 indicating a significant difference.

A descriptive analysis was performed to evaluate the frequency distribution of the demographic variables. Subsequently, a comparison of measurements between the age groups was performed using analysis of variance and post hoc analysis. χ^2 test was used to detect the difference in the presence of broken Shenton line between the groups. χ^2 test was also used to detect any difference in osteochondromas locations over time. Level of significance was set at 0.05. SPSS software was used (version 22; SPSS Inc., Chicago, IL).

RESULTS

Fifty-one children (102 hips) with a diagnosis of HME were included. Of these, 27 (53%) were males and 24 (47%) were females. Mean age at initial presentation was 9.2 years (range, 4.0 to 16.8 y). Four children underwent surgical hip management and only their preoperative radiographs were included in the analysis. Forty-six children (92 hips: 90%) had bilateral distribution of hip osteochondromas. Five children (10 hips, 10%) had no hip lesions on pelvic radiographs at any visit. None of the children had unilateral involvement in our study. A total of 162 AP pelvis radiographs (324 hips) were reviewed (3 children with 6 radiographs; 9 with 5; 10 with 4; 10 with 3; 10 with 2; and 9 children with 1 radiograph).

In the first part of the analysis, when all radiographs were included, femoral neck was the most common location of the osteochondromas (Table 1). There was a tendency toward a better acetabular coverage, measured by Sharp and Wiberg angles, in the older patients (Table 2). More hips with measurements in normal range, of Sharp and Wiberg angles as well as MP, were found in the older age group (Fig. 2). Neck-shaft angle, however, showed some degree of coxa valga that persists in the vast majority of cases (Fig. 2).

In the second part; 18 children (23 hips) had hip subluxation at some point during the study (18/51 children: 35%) (Table 3). Mean age at time of subluxation was 9.4 years (range, 4 to 17 y). No specific location of osteochondromas was found to have a relationship with subluxation (Table 3). The average of measurements at the subluxation diagnosis was: femoral neck-shaft angle 149 degrees, Reimer MP: 33%, Sharp acetabular angle: 50 degrees, Wiberg center-edge angle 13 degrees, and femoral head-neck ratio: 1.

In the third part; 36 children had a minimum follow-up of 2 years (mean follow-up 4.6 y, mean age at first visit 8.5 y and at last visit 13.1 y). In these children, an increased occurrence of lesions was found in almost all locations between the first and the last visits (Table 4). The measurements for the first and last visit were: femoral neck-shaft angle 146.5 and 146.7 degrees, Reimer MP: 23% and 22%, Sharp acetabular angle: 47 and 46 degrees, Wiberg angle 18.9 and 19.8 degrees, and femoral head-neck ratio: 1.1 and 1.2.

In the 5 children (10 hips, 10%) with HME and without hip osteochondromas, mean age at their

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TABLE 2. Comparison of Hip Measurements Between Age Groups

	Mean			Comparison Between Age Groups (*P*)		
Measure	**≤8 y**	**> 8 to < 13 y**	**≥ 13 y**	**≤8 vs. > 8 to < 13 y**	**≤8 vs. ≥ 13 y**	**> 8 to < 13 vs. ≥ 13 y**
Femoral neck-shaft angle	148 deg.	146 deg.	145 deg.	0.028*	< 0.001*	0.112
Reimer migration percentage	25	24	20	0.724	< 0.001*	< 0.001*
Sharp acetabular angle	48 deg.	48 deg.	43 deg.	0.377	< 0.001*	< 0.001*
Wiberg angle	16 deg.	17 deg.	24 deg.	0.118	< 0.001*	< 0.001*
Femoral head-neck ratio	1.09	1.21	1.14	0.224	0.017*	0.517
Shenton line broken, N (%)	49 (48)	44 (43)	34 (28)	0.482	0.021*	0.021*

Analysis of variance and post hoc analysis were used. Level of significance was set as 0.05.
*P value < 0.05 indicating a significant difference.

presentation was 12.1 years (range, 7.7 to 14.3 y). In these children, none of the hips showed a broken Shenton line. Mean radiographic measurements were the following: neck-shaft angle 144 degrees, MP 18%, Sharp angle 44 degrees, Wiberg angle 25 degrees, and femoral head-neck ratio 1.27.

DISCUSSION

In children with HME, osteochondromas can be found in the proximal femur in 77% to 90% of cases.[20,27] In our study, hip osteochondromas were found in 46/51 patients (90%). All these patients with hip osteochondromas had bilateral involvement and their most common location was the femoral neck, followed by the pelvis. These findings support some of the previously reported literature.[14,15,19,28]

More osteochondromas were noted in older patients in most locations (Table 1), demonstrating that these lesions continue to appear during childhood. This pattern was also observed in the longitudinal subgroup of patients who were followed for an average of 4.6 years (Table 4). However, the possibility that the osteochondromas disappear or coalesce with age has been suggested previously.[4,7–10] Although our cross-sectional data (part 1 of the analysis) showed less osteochondromas in the medial femoral neck after age 13 years, this finding could not be confirmed in the longitudinal subgroup. Longer follow-up may be helpful to detect these changes.

In the literature, deformity evolution has been variably described. In 1996, Ozaki et al[16] described a decrease of femoral neck-shaft angle after 10 years of follow-up. In 2001 Malagon[29] described 6 cases, showing that the acetabular dysplasia was caused by the femoral

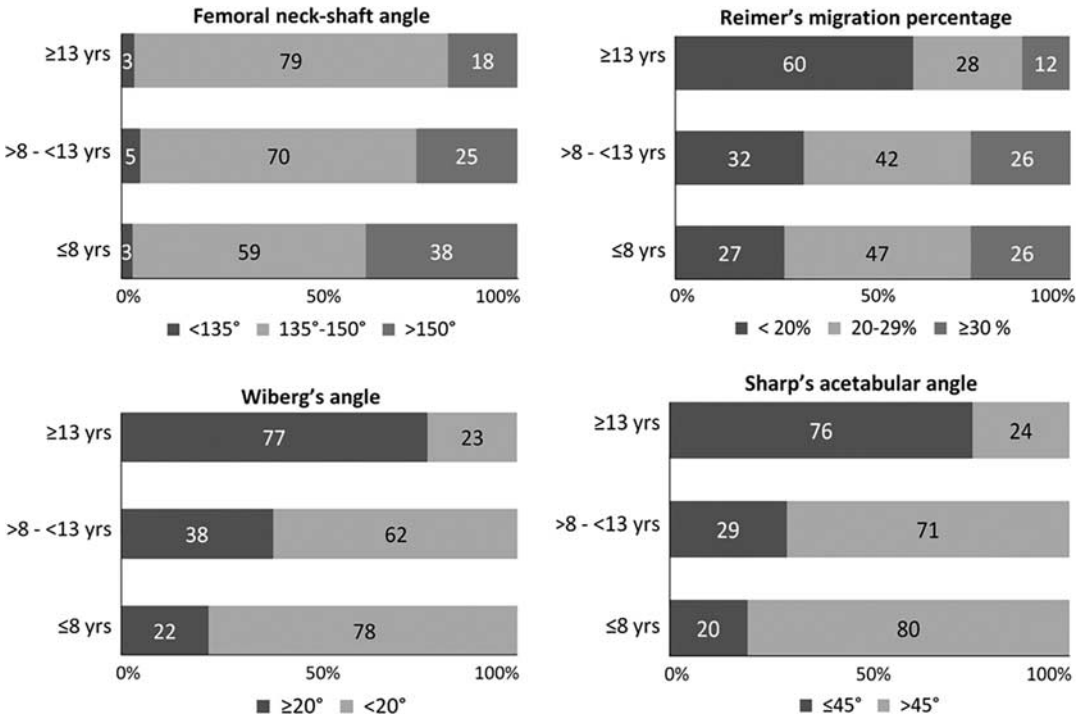

FIGURE 2. Distribution of measurements in the normal range within each age group.

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TABLE 3. Children With Hip Subluxation (MP > 30%) at any Time (23 Hips)

		At Time of Subluxation Diagnosis								Last Hip Radiograph		
			Osteochondromas Locations									
Patient	Hip	Age (y)	MP (%)	MFN	LFN	I	P	A	FH	Age (y)	MP (%)	Time of Follow-up (y)
1	Right	6.8	31		X					9.2	35	2.3
	Left	7.5	31		X					9.2	32	1.7
2	Left	9.6	32		X					NA	NA	0
3	Left	17	31	X	X	X	X			21	26	4
4	Left	9.8	36	X	X	X				11.5	34	1.7
5	Right	4.9	35	X						7.3	36	2.3
6	Right	13.9	32	X	X	X	X			14.7	32	0.8
	Left	12.3	33	X	X			X		14.7	36	2.4
7	Right	6.1	35	X	X	X	X			7	30	0.9
8	Left	16.8	32	X	X	X	X			20	41	3.2
9	Left	12.8	31	X	X	X	X			17.3	26	4.6
10	Right	4.0	39	X					X	6	40	2
11	Right	4.8	33	X	X	X		X		7.7	45	2.9
	Left	4.8	32	X	X	X	X	X		7.7	32	2.9
12	Right	4.8	31	X	X					8.8	35	4
	Left	4.8	33		X					8.8	29	4
13	Right	12	32	X	X	X	X			15.3	27	3.3
	Left	12	32	X	X	X		X		15.3	33	3.3
14	Right	6.6	36	X	X					11.2	38	4.6
15	Right	12.8	32		X					NA	NA	0
16	Right	10.6	38	X	X	X				11.8	42	1.2
17	Right	10.2	40	X	X	X	X			14.5	35	4.3
18	Right	10.8	31	X	X	X				NA	NA	0

A indicates acetabulum; FH, femoral head; I, ischium; LFN, lateral femoral neck; MFN, medial femoral neck; MP, Reimer migration percentage; NA, not applicable; P, pubis; X, osteochondroma noted.

deformity and that these changes were progressive with age. Therefore, surgical excision of the osteochondromas and correction of the deformities were recommended. Later, in 2009, Jellicoe et al[30] reported 2 children with acetabular osteochondromas and acetabular dysplasia. Two years after the excision of osteochondromas, residual acetabular dysplasia was still found. Most recently, a comparison between the measurements, at different ages, was performed by Wang et al,[18] 57 hips (30 patients) were studied and showed an increase in the MP over time with a decrease of femoral neck-shaft angle and acetabular index. In this study, normal hip measurements in the normal range were recorded more often in the older age group (> 13 y) (Table 2 and Fig. 2). Although individual improvement, in radiographic measurements, was noted (Figs. 3A, B); the longitudinal group did not show any change in the average radiographic measurements over follow-up; however, the mean age at last visit was 13 years and changes after this age could not be assessed. Sharp acetabular angle uses the teardrop figure and it is generally used after the triradiate cartilage is fused. However, the teardrop starts to be visible during childhood and therefore, it can be used as an important parameter in the evaluation of hip development.[31] Many previous studies, some of them about HME, used the teardrop and Sharp acetabular angle in pediatric population.[14,15,32]

TABLE 4. Comparison of Osteochondromas Locations Between the First and Last Radiographs in Children With Minimum 2 Years of Follow-up (Total 72 Hip Radiographs)

Location	First X-ray Visit, N (%)	Last X-ray Visit, N (%)	*P*
Medial femoral neck	37 (51)	53 (74)	0.006*
Lateral femoral neck	35 (49)	35 (49)	1.000
Ischium	24 (33)	45 (63)	0.0005*
Pubis	17 (24)	21 (29)	0.449
Acetabulum	9 (13)	17 (24)	0.083
Femoral head	0 (0)	2 (3)	0.154

χ^2 test was used.
*P value < 0.05 indicating a significant difference.

Hip subluxation, at any time of the study period, was noted in 23% of the hips (23/102 hips). Others studies reported prevalence of hip subluxation between 5% and 19%.[15,18] Similar to our study, none of the prior studies found any relationship between hip subluxation and the location of osteochondromas.

In the 5 children with HME and no hip osteochondromas, coxa valga was found on radiographs supporting previous reports.[14,18,18,33] We could not find a relationship between coxa valga and hip dysplasia in this study. These findings are similar to those of Porter et al[14] who also could not find a relationship between these 2 deformities.

In addition to the retrospective nature, limitations of our study include the number of patients, the

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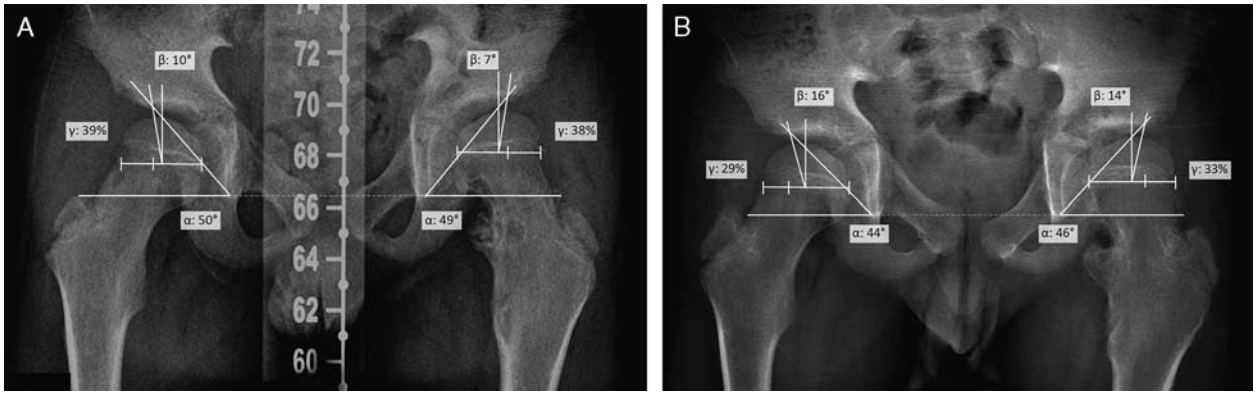

FIGURE 3. A, Anteroposterior pelvis radiograph of a 9.8-year-old boy with hereditary multiple exostoses. B, Anteroposterior pelvis radiograph of the same patient at 12.4 years of age. The increase of Wiberg angle (β) and the decrease of Sharp angle (α) and Reimer migration percentage (γ) are shown.

cross-sectional nature of the analysis in the first part and the follow-up term for the longitudinal subgroup. Only AP pelvis radiographs were used for osteochondromas detection due to the availability of these radiographs for all patients. Although missing a radiolucent lesion might have been possible in this study,[15,34] all radiographs were measured by 1 reviewer using the same methods. Reliability of the measurements was not assessed in this study as it was well documented in previous studies and as all the radiographs were measured by a single observer.

Individual improvement in radiographic measurements were noted in our study; however, the prevalence of hip osteochondromas in patients with HME was high (90%), hip subluxation was found in 23% of cases and even in children with no hip osteochondromas, radiographic abnormalities were found. We do feel that having a baseline set of radiographs and another set at skeletal maturity are important as these patients are at risk for malignant transformation and the pelvis is a relatively common site for this. We do not routinely get surveillance radiographs at each visit. Surveillance is suggested only in patients with radiographic abnormalities such as subluxation.

This study suggests that, in children with HME, radiographic evaluation of the hip is necessary based on the high percentage of hip involvement. Radiographic surveillance should be considered to monitor hip subluxation. In a previous study,[13] a pelvis radiograph was recommended at the time of HME diagnosis for the early detection of hip osteochondromas. However, there is still no consensus regarding this recommendation in the literature. We agree with having a pelvis radiograph at the time of HME diagnosis with later radiographic follow-up to be determined based on the presence of pelvic osteochondromas. Surgery may certainly be necessary for symptomatic osteochondromas. However, given the possibility of improvement in hip parameters with age, early surgical treatment to improve hip longevity does not seem to be warranted.

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