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Title

Osteoarthritis of the Hip in Fibrous Dysplasia of the Proximal Femur

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Abstract

Background: Proximal part of the femur is one of the most common site of fibrous dysplasia (FD). The strong mechanical forces of the proximal femur on the weakened dysplastic bone often results in deformity of the proximal femur in FD patients. The deformity of the proximal femur or coxa vara results in the decrease of the neck-shaft angle, and is often associated with functional impairment and pain in FD patients.

Coxa vara deviates the normal mechanical axis and may predispose the hip joint to secondary osteoarthritis (OA). Understanding the prevalence and predisposing factors of hip OA is necessary to provide the guidelines as to the treatment planning and surveillance for FD patients. However, as far as we know, no study has been reported with regard to hip OA in FD of proximal femur.

Questions/Purposes: This study investigated the prevalence and associated factors for hip OA in FD of the proximal femur.

Patients and Methods: From the prospectively collected outpatient database of our department, 370 consecutive patients with the diagnosis of FD between 2008 and 2014 were identified. Of the 370 patients, 139 patients without hip radiographs were excluded, 10 patients without FD of the proximal femur, 12 patients with incomplete medical records were additionally excluded, leaving 209 patients for inclusion in the study.

The presence of OA was determined using the anteroposterior radiograph of the hip using the Kellegren-Lawrence (KL) grading system. We defined radiological hip OA as a KL grade of 1 or higher. The interobserver agreement was substantial (k coefficient value = 0.83).⁹ If there was a discrepancy between the observers, a consensus agreement was reached.

Medical records were reviewed for the potential radiological and clinical factors that might be associated with the presence of OA in FD of proximal femur: (1) patient demographics, (2) deformity of the proximal femur

(coxa vara), (3) peri-articular location of the tumor, (4) skeletal disease burden, and (5) presence of endocrine abnormalities. There were 91 men and 118 women, and median age at the time of last hip radiographs was 38 years (range, 6-74 years). Fifty-eight (29%) of 209 patients had previously undergone surgery for pain or deformity due to FD of the proximal femur. The degree of deformity of the proximal femur was determined by measuring the neck shaft angle. The mean neck-shaft angle was 127 degrees (range, 64 - 148). Peri-articular location of the tumor was examined as to the local presence of the tumor in the peri-articular bones; acetabulum or femoral head. There were 35 cases (17%) involving the femoral head and 43 cases (21%) involving the acetabulum. Skeletal disease burden was measured by quantifying the number of bones involved on bone scan (n=50) or plain radiographs (n=159). There were 148 (71%) monostotic types and 61 (29%) polyostotic types. The average number of bones involved for polyostotic types was 4 (range, 2-15). Among the 61 patients with polyostotic type FD, 4 patients were identified with endocrinopathies.

Results: Of the 209 patients, 24 (12%) had radiographic evidence of hip OA. 17 patients had KL grade 1 OA (71%), 5 patients KL grade 2 OA (21%) and 2 patients had KL grade 3 OA (8%). The average age at diagnosis of hip OA was 32 years (range, 14-59). Of the 24 patients with hip OA, only 1 out of 148 patients (1%) in the monostotic group had OA whereas 23 of 61 patients (38%) showed OA in the polyostotic group ($p<0.001$).

When characteristics of the patients with hip OA (n=24) were compared those of the patients without hip OA (n=185), the proportion of polyostotic types was significantly higher in the patients with hip OA ($p<0.001$) (Table 1). To further analyze the associated factors for hip OA, only the patients with polyostotic types were selected. On univariate logistic regression analysis of associated factors of hip OA in polyostotic type, neck shaft angle ($p<0.001$), presence the tumor in the femoral head ($p<0.001$) and presence of the tumor in the acetabulum ($p<0.001$) were significant (Table 2). On multivariate analysis, neck shaft angle (HR=0.847 per 1° increase, $p=0.004$), presence of femoral head lesion (HR=9.947, $p=0.027$) and presence of acetabular lesion (HR=11.231, $p=0.014$) remained as independent factors associated with hip OA in polyostotic type of proximal femur FD.

Conclusions: Our data suggest that the risk of developing hip OA is considerable in polyostotic type of FD involving the proximal femur. Development of hip OA was influenced by the presence of femoral head and acetabular lesion and the degree of deformity in FD of proximal femur.

Table 1. Comparison between patients with and without hip osteoarthritis

Characteristic	With osteoarthritis (n=24)	Without osteoarthritis (n=185)	P value
Age (years)	31.5 (\pm 12.9)	39.1 (\pm 17.0)	0.036
Sex			0.577
Female	15 (62%)	103 (56%)	
Male	9 (38%)	82 (44%)	
Neck shaft angle (°)	111.7 (\pm 14.7)	128.5 (\pm 7.3)	<0.001
Type			<0.001
Monostotic	1 (4%)	147 (82%)	
Polyostotic	23 (96%)	33 (18%)	
Number of bones involved	4.8 (\pm 3.6)	1.5 (\pm 1.4)	<0.001
Femoral head lesion			<0.001
Absent	9 (38%)	165 (89%)	
Present	15 (62%)	20 (11%)	
Acetabular lesion			<0.001
Absent	3 (13%)	163 (88%)	
Present	21 (87%)	22 (12%)	
Endocrine abnormality			0.054
Absent	22 (92%)	183 (99%)	
Present	2 (8%)	2 (1%)	

Table 2. Factors associated with hip osteoarthritis in polyostotic type fibrous dysplasia

Factors	Univariate			Multivariate		
	HR	95%CI	P	HR	95%CI	P
Age	0.990	0.955-1.027	0.602			
Sex			0.445			
Male (n=25)	1					
Female (n=36)	1.518	0.521-4.426				
Neck-shaft angle (°)	0.878	0.818-0.941	<0.001	0.847	0.757-0.947	0.004
Number of bones involved	1.203	0.988-1.464	0.066			
Femoral head lesion			<0.001			0.027
Absent (n=38)	1			1		
Present (n=23)	7.031	2.205-22.419		9.947	1.306-75.757	
Acetabular lesion			<0.001			0.014
Absent (n=28)	1			1		
Present (n=33)	12.821	3.205-51.820		11.231	1.647-76.582	
Endocrine abnormality			0.590			
Absent (n=55)	1					
Present (n=4)	1.750	0.229-13.398				