Exostosis-Like Intra-articular Periosteal Osteoblastoma: A Rare Case

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Abstract

Osteoblastoma is a relatively rare benign bone tumor, most often located in the vertebral column or metaphysis of the long bones, particularly the femur and the tibia. Periosteal osteoblastoma is the least common type. Exostosis-like appearance is not common even in periosteal osteoblastoma, a very rare type of this tumor. In addition, an intra-articular location is uncommon for osteoblastomas.

Here we report the case of a 25-year-old man with intra-articular exostosis-like periosteal osteoblastoma of the hip that resulted in impingement and osteoarthritis.

Osteoblastoma is an uncommon benign bone-forming tumor that accounts for approximately 3.5% of benign neoplasms and less than 1% of all bone neoplasms.[1] Most patients are 10 to 30 years old, and men outnumber women by a factor of 2 or 3 to 1.[2] The most common site for this tumor is the posterior part of the spine, then the metaphysis of the long bones, particularly the femur and the tibia.[2]

Most patients present with pain,[1] though swelling may be the presenting sign in subcutaneous bones.[2] Pain may be similar to what is experienced with osteoid osteoma—namely, nocturnal pain that responds dramatically to nonsteroidal anti-inflammatory drugs (NSAIDs). Unfortunately, this is not the response most of the time.[2]

Most femoral osteoblastomas occur in the trochanteric area, and head and neck involvement is extremely rare.[1-3] In addition, a periosteal origin for an osteoblastoma is not common; only 29 cases have been reported.[4,5] In this article, we report a case of periosteal osteoblastoma with a large protruding mass on the intra-articular part of the femoral neck leading to hip osteoarthritis. The patient provided written informed consent for print and electronic publication of this case report.

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Case Report

A 25-year-old otherwise healthy man presented to our clinic with the chief complaint of a 3-year history of right hip pain that started dramatically worsening 6 months before presentation. The pain was aggravated by activity but was also present at night, and it responded well to NSAIDs, particularly early in its course. The patient had a history of ipsilateral hip trauma 8 years earlier. The trauma had been relatively minor, and the pain had subsided over a few days without treatment. The patient had a severe limp, global limitation of motion, and abduction and flexion contracture.

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Radiograph of the pelvis showed moderately severe osteoarthritis of the hip along with a bony protuberance on the inferior aspect of the most proximal part of the neck, near its junction with the head (Figure 1). Underneath, a continuous layer of cortical bone was seen, though slight saucerization of the cortex was evident. The medullary canal was not involved.

T_2-weighted magnetic resonance imaging (MRI) showed a hyperintense lesion bulging out from the inferior surface of the cortex of the femoral neck (Figure 2). Fat-suppression MRI showed edema of the interior of the lesion (Figure 3). Other MRI findings were destruction of the articular cartilage and joint effusion. Differential diagnosis included benign bone tumors (eg, osteochondroma, osteoblastoma) and traumatic osteochondral fragment. Traumatic osteochondral fragment

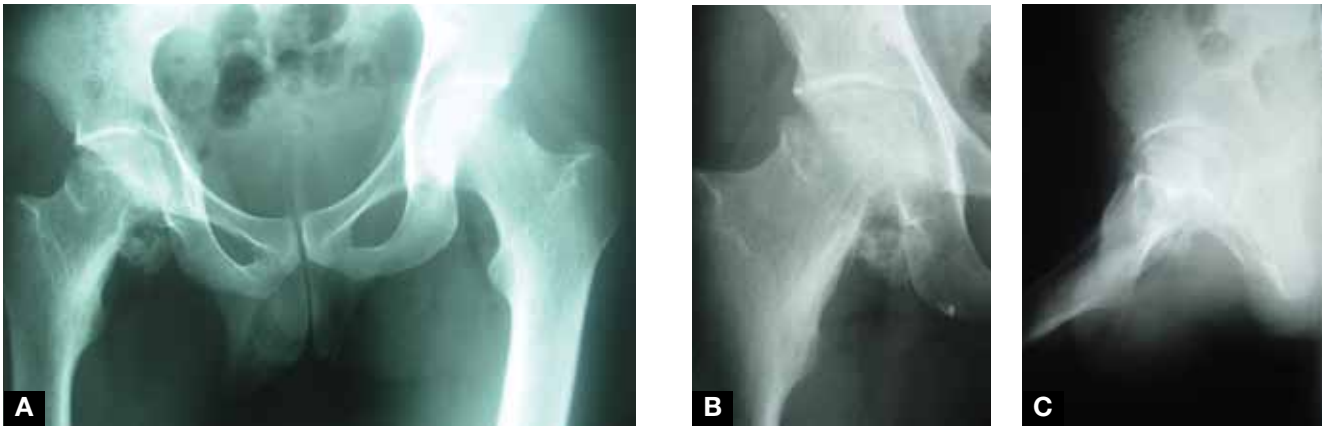

Figure 1. Prominent, exostosis-like periosteal osteoblastoma at inferior border of head–neck junction. (A) Anteroposterior view of pelvis. (B) Anteroposterior view of right hip. (C) Lateral view of right hip.

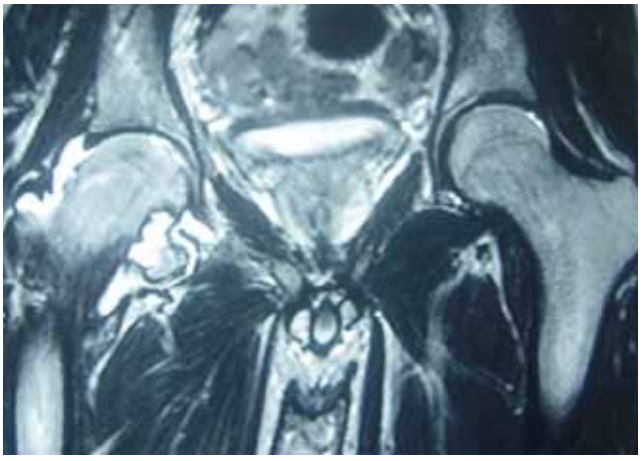

Figure 2. T_2-weighted coronal magnetic resonance imaging of pelvis. Note edematous prominent lesion at inferior border of head–neck junction.

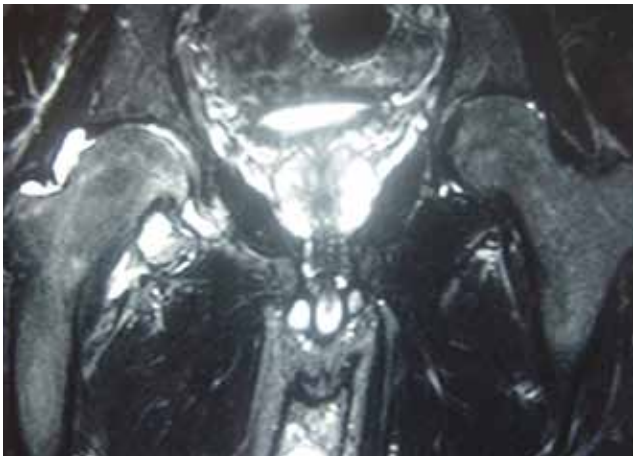

Figure 3. Fat-suppression coronal magnetic resonance imaging of pelvis. Note hyperintensity signal of intra-articular effusion.

was not a strong possibility given the inconsistent history and the fact that the original site from which such a large fragment would have been detached was not visible on radiograph or MRI. Osteochondroma was also improbable given the lack of cortical continuity of the tumor and the femoral neck. The most probable diagnosis, though rare, was periosteal osteoblastoma. The patient was nominated to undergo total hip arthroplasty to treat both the coxarthrosis and the bony lesion.

Hip arthrotomy was performed through the direct lateral approach of Hardinge. The lesion was easily visible on the inferior border of the neck. The entire neck, including the bulging lesion, was removed with a neck cut just as is routinely done in conventional arthroplasty. Joint replacement was performed uneventfully.

On gross pathologic examination of the specimen, a segment of femoral head and neck was found along with the bony prominence, measuring in total 4.5×3×2 cm. Histologic examination of the lesion showed an interlacing network of bone trabeculation evenly distributed in a loose fibroblastic stroma with prominent vasculature. An obvious layer of rimming osteoblasts and multinucleated osteoclast-like giant cells was present. In addition, an inflamed and edematous synovium with foci of lymphoid follicles was present (Figure 4).

Discussion

Intra-articular, periosteal, prominent osteoblastoma is very rare. To our knowledge, it was reported only once in the English-language literature.[3] The proximal femur is the most common site of involvement after the spine, but it is very rare for an osteoblastoma to arise proximal to the intertrochanteric line.[1,2,5]

The intra-articular part of the proximal femur is a relatively common site for osteoid osteoma.[6-8] This is not the case with osteoblastoma. Several cases of intra-articular medullary osteoblastoma have been reported, but all were confined to the boundaries of the femoral neck cortex. To our knowledge, only one case of intra-articular osteoblastoma directly in contact with joint fluid has been reported in the English-language literature, and it was also a periosteal osteoblastoma located on the distal femoral neck.[3]

Regarding the anatomical site of origin, osteoblastoma may be medullary, intracortical, or periosteal.

Table. Reported Cases of Periosteal Osteoblastoma

Location	Age (y)	Sex	Year	Reported By
Humerus	7	F	1964	Lichtenstein & Sawyer[2]
Femoral diaphysis	—	—	1964	Lichtenstein & Sawyer[2]
Greater trochanter of femur	10	F	1971	Goldman[10]
Distal anterior tibia	28	M	1971	Goldman[10]
Mandible	9	M	1976	Farman et al[11]
Maxilla	30	F	1978	Chatterji et al[12]
Femoral neck	17	M	1982	Tonai et al[3]
Femoral diaphysis	51	M	1982	Tonai et al[3]
Fifth rib	19	M	1989	Gentry et al[13]
Proximal humerus	29	—	1989	Mirra et al[14]
Seventh rib	32	M	1991	Huvos[15]
Upper third of humerus	19	M	1994	Schajowicz[16]
Upper third of radius	30	M	1994	Schajowicz[16]
Ninth rib	23	F	1994	Schajowicz[16]
Frontal bone	—	—	1994	Schajowicz[16]
Occipital bone	—	—	1994	Schajowicz[16]
Jaw	—	—	1994	Schajowicz[16]
Upper third of fibula	16	M	1994	Schajowicz[16]
Fibula	—	—	1994	Schajowicz[16]
Proximal tibial metaphysis	22	M	1994	Schajowicz[16]
Femur	18	M	1994	Schajowicz[16]
Scapula	39	M	1994	Schajowicz[16]
Distal femur	—	—	1997	Forest et al[17]
Distal humerus	12	M	1998	Kawaguchi et al[18]
Posterior distal femoral shaft	24	M	2000	Sulzbacher et al[19]
Posterior distal femoral shaft	17	M	2004	Nakatani et al[20]
Ethmoid bone	66	M	2004	Lee et al[21]
Frontal bone	32	F	2005	Lin et al[22]
Proximal tibial metaphysis	20	F	2007	Mortazavi et al[5]

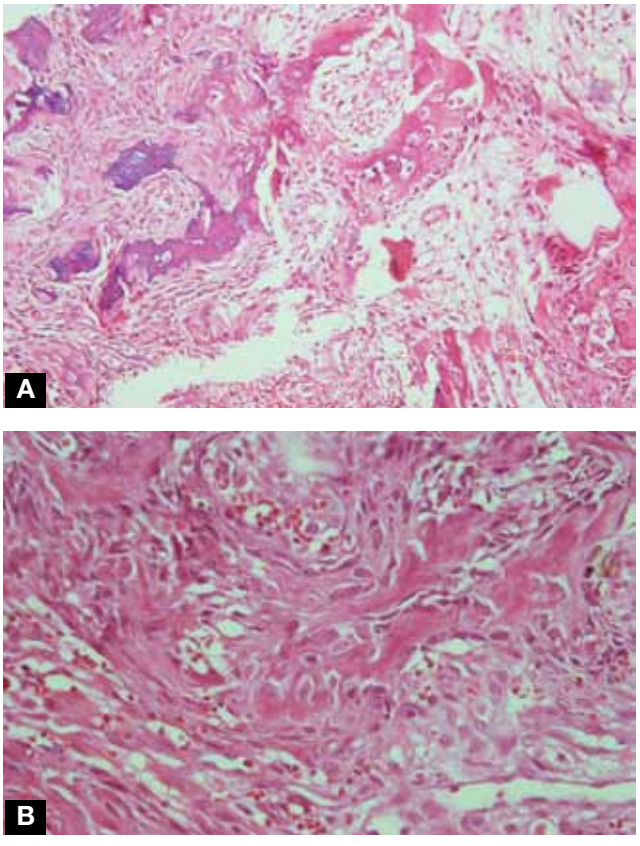

Figure 4. (A) Typical microscopic view of osteoblastoma with rimming osteoblasts, giant cells, and bone trabeculations in vascular stroma. (B) High-power microscopic view of nidus containing irregular trabecula of osteoid bordered by osteoblasts set in a vascular stroma.

Medullary and intracortical osteoblastomas are the most common, periosteal the least.[1,9] A lesion that originates under the cortex is considered a medullary osteoblastoma; although there may be cortical expansion with this lesion, exostosis-like protrusion through the cortex is not expected. Twenty-nine cases[2,3,5,10-22] (Table) of periosteal osteoblastoma have been reported.[5] Most often, they do not show marked bulging, and only seldom do they appear pedunculated. The present case, an obvious periosteal osteoblastoma, was clearly prominent and appeared in an unusual location. Intracortical lesions differ from our patient's lesion in that they never protrude from the cortex.[2,9] Osteoarthritis of the affected hip was attributed to tumor mass impingement of the acetabulum, which was confirmed by the marked abduction contracture. The mass was large and could easily come into contact with the acetabular rim during motion, adduction in particular. It seems logical that, if the bony prominence had been excised early in the course, coxarthrosis could have been prevented.

Hip arthritis manifested by effusion (MRI) and confirmed by pathologic study was probably caused by the tumor and by its secondary effects on the articular cartilage. There is evidence that secretion of prostaglandins and other inflammatory mediators may be the main cause of joint inflammation in intra-articular osteoid osteoma.[23] Given the similarities between osteoblastoma and osteoid osteoma, the same mechanism may have been at work in the present case.

Regarding the histopathologic features of the mass, the present case was a typical osteoblastoma with

bony trabeculae lined by a single layer of osteoblasts. Although uncommon, such a radiologic manifestation for an osteoblastoma is possible, and periosteal osteoblastoma should be considered in the differential diagnosis of an exostosis-like lesion, particularly when the medullary canal of the lesion is not continuous with that of the underlying bone. In addition, a surface osteoblastoma can cause joint osteoarthritis and should be included in the differential diagnosis of apparently idiopathic arthritis.

Authors' Disclosure Statement

The authors report no actual or potential conflict of interest in relation to this article.

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This paper will be judged for the Resident Writer's Award.