American Journal of Case Reports

ISSN 1507-6164
© Am J Case Rep, 2013; 14: 258-262
DOI: 10.12659/AJCR.889243

Received: 2013.04.02
Accepted: 2013.05.09
Published: 2013.07.17

Intraarticular osteoblastoma with subluxation of the hip joint

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

ABDE 1 Kyoji Okada
BCF 2 Hiroyuki Nagasawa
BDF 2 Schuichi Chida
AD 3 Hiroshi Nanjo

1 Department of Physical Therapy,Akita University Graduate School of Health Science, Hondo, Akita, Japan
2 Department of Orthopedic Surgery, Akita University Graduate School of Medicine, Hondo, Akita, Japan
3 Department of Clinical Pathology, Akita University Hospital, Hondo, Akita, Japan

Corresponding Author: Kyoji Okada, e-mail: cshokada@med.akita-u.ac.jp

Patient: Male, 5
Final Diagnosis: Osteoblastoma
Symptoms: —
Medication: —
Clinical Procedure: —
Specialty: Oncology

Objective: Rare disease
Background: Osteoblastomas are relatively uncommon bone tumors that account for <1% of all bone tumors. They usually occur in the medullary region of the bone. As such, intraarticular osteoblastomas are quite rare.
Case Report: In this report, we present the case of a 5-year-old boy who presented with vague pain and subluxation of the hip joint due to an intraarticular osteoblastoma. Radiological examinations showed an irregular calcified mass lesion in the hip joint. The final diagnosis of osteoblastoma was made by histological examination. The patient's symptoms completely subsided following surgical removal of the tumor.
Conclusions: Osteoblastomas can occur in the intraarticular region. Although quite rare, osteoblastoma should be considered among the differential diagnoses for patients with pain and subluxation of the hip joint.

Key words: intraarticular • diagnosis • bone neoplasms • osteoblastoma

Full-text PDF: http://www.amjcaserep.com/download/index/idArt/889243

Background

Osteoblastomas are bone-forming benign tumors that account for <1% of all bone tumors and are usually larger than osteoid osteomas [1]. The lesions may originate in any skeletal location but most commonly occur in the posterior part of the spine, followed by the appendicular skeleton. Most osteoblastomas occur in the medullary region of the bone and rarely occur on the surface of the bone [2]. Among osteoblastomas arising on the surface of the bone, intraarticular conditions have been quite rare [3,4]. In this study, we report only the third case of an intraarticular osteoblastoma associated with subluxation of the hip joint.

Case Report

In December 2003, a 5-year-old boy was referred to our institute for the evaluation of left hip pain that had restricted ambulation for several months. On physical examination, the left hip showed contracture of abduction and external rotation, with a decrease in the flexion, adduction, and internal rotation of the hip. The boy complained of vague pain on passive motion of the hip joint. Mild tenderness was found around the joint, but no local heat was detected. Body temperature was normal, and laboratory data was unremarkable.

Radiographs revealed a lateral subluxation of the left hip joint. Tomography demonstrated a mineralized lesion adjacent to the surface of the joint. Computed tomography (CT) scan revealed an intraarticular lesion with erratically distributed mineralization. The lesion was 2×3 cm in size and located adjacent to the acetabulum (Figure 1). The lesion showed a low signal intensity on T1-weighted magnetic resonance (MR) images and relatively high signal intensity on T2-weighted images. Joint effusion around the lesion was observed on the T2-weighted images. The acetabulum adjacent to the lesion showed diffuse low signal intensity on T1-weighted MR images, but these signal changes were not observed on T2-weighted MR images (Figure 2).

An excisional biopsy was performed using the anterior approach. After capsulotomy, a brown-colored, irregularly mineralized, friable lesion was found attached to the acetabulum immediately inferior to the triradiate cartilage. The lesion was 3 cm in diameter at its largest point. The intraoperative pathological diagnosis on the frozen section procedure was a benign bone-forming tumor. The lesion was completely excised. The acetabulum, joint cartilage, and the round ligament appeared normal. Mild proliferation of the synovium was also observed, but no apparent relationship between the lesion and the synovium was observed. On histological examination of the formalin-fixed, paraffin-embedded tissue, the lesion showed a proliferation of osteoblasts with an abundant formation of bony trabeculae. There was mild nuclear atypia of these osteoblasts without atypical mitosis. Proliferation of vessels was also observed around the bony trabeculae. Based on these findings, a final diagnosis of osteoblastoma was made (Figure 3).

One day after the surgery, the boy did not experience pain during passive movements of the leg. We permitted weight bearing 3 weeks after the surgery. At an 8-year postoperative follow-up, he continued to be asymptomatic, and radiographs showed no abnormality around the left hip joint.

Discussion

Most osteoblastomas occur in the medullary regions of the bone and rarely occur on the surface of the bone. In the largest noted

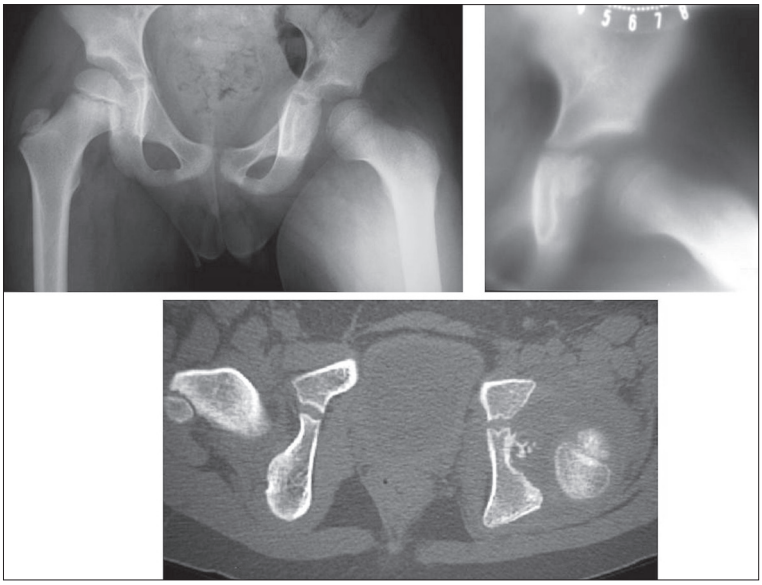

Figure 1. Upper left: Radiograph showing a lateral subluxation of the left hip joint. Upper right: Tomograph taken at admission showing a mineralized lesion adjacent to the joint surface of the joint. Lower: Computed tomography scan demonstrating the intraarticular lesion with erratically distributed mineralization. The lesion was 2×3 cm, and was located adjacent to the acetabulum.

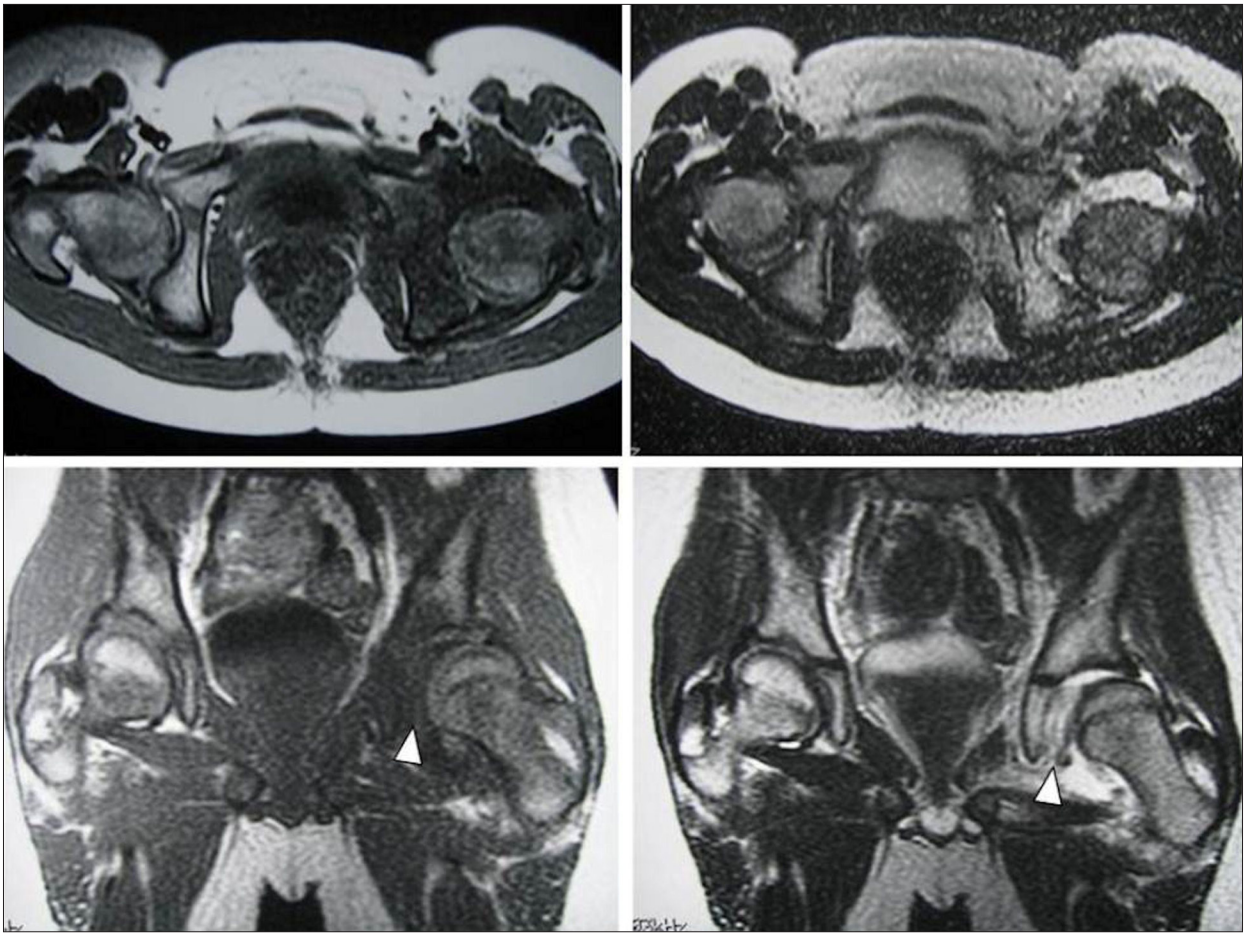

Figure 2. Upper left, lower left: T1-weighted magnetic resonance image showing low-signal intensity in the lesion (white arrow head). The acetabulum adjacent to the lesion shows diffuse low signal intensity. Upper right, lower right: The lesion shows a high signal intensity on T2-weighted images (white arrow head). Joint effusion around the lesion is also observed. Signal changes in the acetabulum observed on T1-weighted images are not apparent on T2-weighted magnetic resonance images.

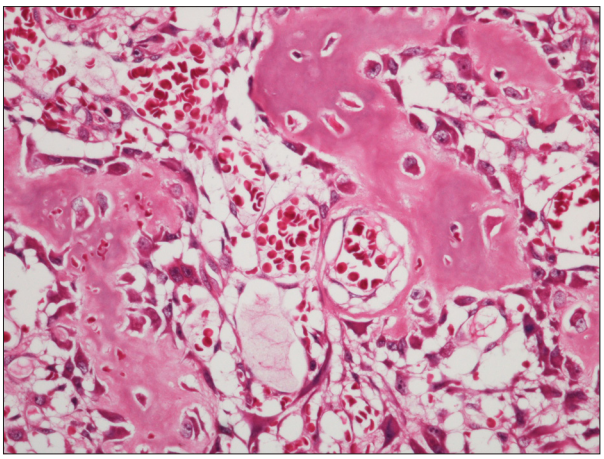

Figure 3. Microphotograph of the lesion showing a proliferation of osteoblasts with an abundant formation of bony trabeculae. Nuclear atypia of these osteoblasts is mild, and no atypical mitosis is observed (Hematoxylin and eosin stain, original magnification, ×400.

series of osteoblastomas, only 4% occurring in appendicular skeletons were surface-type [2]. According to Abolghasemian et al., in 2010, 29 surface-type osteoblastomas were reported, most of which were diagnosed as periosteal osteoblastomas [4]. In the current case, the medullary region of the acetabulum that was adjacent to the lesion showed diffuse low signal intensity on T1-weighted MR images; however, intraoperative appearances and 8-year follow-up confirmed that there was no neoplastic change in that region. Therefore, the current case should be classified as a surface-type osteoblastoma.

Within surface-type bone tumors, intraarticular lesions are a peculiar subtype. In osteoid osteomas, an intraarticular location is not uncommon [1,5], while intraarticular osteoblastomas are quite rare. To our knowledge, only 3 intraarticular osteoblastomas (including the current case) have been reported in the English literature [3,4]. Tonai et al. reported the first case of a 17-year-old boy with an intraarticular osteoblastoma arising from the surface of the femoral neck in 1982 [3].

Table 1. Literature review of cases with intraarticular osteoblastoma.

No.	Authors	Year	Age (year)	Sex	Primary Site	Symptoms
1	Tonai	1982	17	M	Femoral neck	Pain, limited ROM
2	Abolghasemian	2010	25	M	Femoral neck	Pain, limited ROM
3	Current case	2013	5	M	Acetabulum	Pain, limited ROM

ROM – range of motion.

In 2010, Abolghsemian et al. reported a 25-year-old man with an intraarticular osteoblastoma arising from the proximal femur[4]. Several characteristics were consistent in all the 3 cases. All the patients were male, with a relatively young age of onset. Furthermore, all lesions involved the hip joint, and all patients complained of a vague hip pain associated with limitation of motion (Table 1). Rarely, osteoblastoma arising in the acetabulum could extend into the hip joint [6].

Two of the 3 cases reported associated MR image findings. Both cases showed joint effusion of the affected joint, and the current case showed a non-neoplastic signal change of the acetabulum adjacent to the neoplasm. In osteoid osteoma, osteoblastoma and chondroblastoma, neoplastic production of prostaglandin or other chemical mediators have been considered to be associated with synovitis of adjacent joints and signal changes of surrounding tissues in MR images [7–9]. In the current case, these mediators may be a cause of synovitis and the signal change in surrounding tissues.

In the current case, lateral subluxation of the hip joint was a characteristic. In synovial chondromatosis of hip joints, widening of the joint space is a common feature. Yoon et al. reported that 12 of the 21 cases with synovial chondromatosis of the hip joint showed widening of the joint space [10]. Rarely, subluxation of the hip joint associated with synovial chondromatosis has been reported [11]. Therefore, synovial chondromatosis should be included in the differential diagnosis. Usually, dotted or round regular mineralization patterns are observed in synovial chondromatosis. In contrast, mineralization patterns of osteoblastomas are irregular and variable. In the current case, tomography and CT scans showed irregular distributed mineralization of the lesion. These are unlikely to occur in synovial chondromatosis. Furthermore, metaplastic cartilage proliferation is a histological finding of synovial chondromatosis. Diffuse bone forming features in the current case were not compatible with those of synovial chondromatosis.

Other bone forming neoplasms were included in the differential diagnosis. Since intraarticular osteoid osteoma could cause hip subluxation, it should also be included in the differential diagnosis [12]. Osteoblastomas and osteoid osteomas are both benign bone-forming neoplasms with identical histological features; however, osteoid osteomas usually have a small nidus measuring, <1 cm, while the nidus of osteoblastomas tend to be larger [1]. Since the current lesion measure 3 cm at the largest diameter, osteoblastoma was deemed the appropriate diagnosis.

Osteosarcomas occur on the surface of the bone [13,14]. However, osteosarcomas that are restricted to joint space have not been reported previously. Osteosarcomas restricted to the hip joint are not likely as a diagnosis of the current case, although histological evaluation of a biopsied specimen or excisional material is mandatory to confirm the final diagnosis.

Conclusions

Osteoblastomas can occur as intraarticular lesions. To the best of our knowledge, this is only the third reported case of an intraarticular osteoblastoma. Although quite rare, osteoblastoma should be considered among the differential diagnoses for patients with pain and subluxation of the hip joint.

References:

1. Unni KK, Inwards CY: Osteoblastoma (giant osteoid osteoma). In: Dahlin's bone tumors: general aspects and data on 10,165 cases. 6th ed. Philadelphia: Lippincott Williams & Wilkins, 2010; 112–21
2. Lucas DR, Unni KK, McLeod RA et al: Osteoblastoma: clinicopathologic study of 306 cases. Hum Pathol, 1994; 25: 117–34
3. Tonai M, Campbell CJ, Ahn GH et al: Osteoblastoma: classification and report of 16 patients. Clin Orthop Relat Res, 1982; 167: 222–35
4. Abolghasemian M, Rezaie M, Behgoo A et al: Exostosis-like intra-articular periosteal osteoblastoma: a rare case. Am J Orthop, 2010; 39: E50–53
5. Szendroi M, Kollo K, Abtal I et al: Intraarticular osteoid osteoma: clinical features, imaging results, and comparison with extraarticular localization. J Rheumatol, 2004; 31: 957–64
6. Arauz S, Morcuende JA, Weinstein SL: Intra-articular benign osteoblastoma of the acetabulum: a case report. J Pediatr Orthop B, 1999; 8: 136–38
7. Makley JT, Dunn MJ: Prostaglandin synthesis by osteoid osteoma. Lancet, 1982; 2: 43
8. Yamamura S, Sato K, Sugiura H et al: Prostaglandin levels of primary bone tumor tissues correlated with peritumoral edema demonstrated by magnetic resonance imaging. Cancer, 1997; 79: 255–61

9. James SIJ, Panicek DM, Davies AM: Bone marrow oedema associated with benign abd malignant bone tumours. Eur J Radiol, 2008; 67: 11–21
10. Yoon PW, Yoo JJ, Koo K-H et al: Joint space widening in synovial chondromatosis of the hip. J Bone Joint Surg, 2011; 93A: 303–10
11. Hardacker J, Mindell E: Synovial chondromatosis with secondary subluxation of the hip. J Bone Joint Surg, 1991; 73A: 1405–7
12. Ninomiya S, Mamamda K, Ozawa H: Subluxation of the hip from osteoid osteoma–Report of 2 intraarticular cases. Acta Orthop Scand, 1989; 60: 220–22
13. Okada K, Frassica FJ, Sim FH et al: Parosteal osteosarcoma. A clinicopathological study. J Bone Joint Surg, 1994; 76A: 366–78
14. Okada K, Unni KK, Swee RG, Sim FH: High-grade surface osteosarcoma. Cancer, 1999; 85: 1044–54