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Case Report

Polyostotic osteoid osteoma: A case report

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ABSTRACT

Osteoid osteomas are common, benign osteoblastic tumors that can occur in any bone in the body. They are almost always solitary, with only rare reports of multiple tumors in the same patient. When multiple, they typically are found within the same bone. We present a unique case of a young female athlete who presented initially at 16 years old with a right tibial osteoid osteoma and later at 18 years old with a right acetabular osteoid osteoma. Our case demonstrates the rare entity of polyostotic osteoid osteoma, the potential limitations of MRI in the diagnosis of osteoid osteoma, and the utility of radiofrequency ablation in the treatment of osteoid osteoma.

Introduction

Osteoid osteoma is a benign osteoblastic bone tumor first described by Jaffe in 1935 [1]. It consists of a highly vascular central nidus made of osteoid and woven bone with a fibrovascular rim and surrounding reactive bone formation [2,3]. Classically, osteoid osteomas are associated with progressive pain that is worse at night and relieved with salicylates. Osteoid osteomas comprise approximately 12% of all benign skeletal neoplasms and most commonly affect adolescents and young adults in a 2:1 male to female ratio [2,3].

The classic radiographic appearance is an oval, radiolucent central focus smaller than 2 cm with surrounding reactive sclerosis [4,5]. CT is the modality of choice for diagnosis as the dense central sclerotic focus and radiolucent rim may be obscured on radiography and MRI.

Osteoid osteomas typically occur in the cortex of long bones of the lower extremities but can occur in any bone and can be intramedullary or subperiosteal [2]. They are almost always solitary, as multiple osteoid osteomas within the same patient comprise less than 1% of reported cases [6]. When multiple, osteoid osteomas typically occur as nidi in the same bone rather than tumors in separate bones [7]. We present

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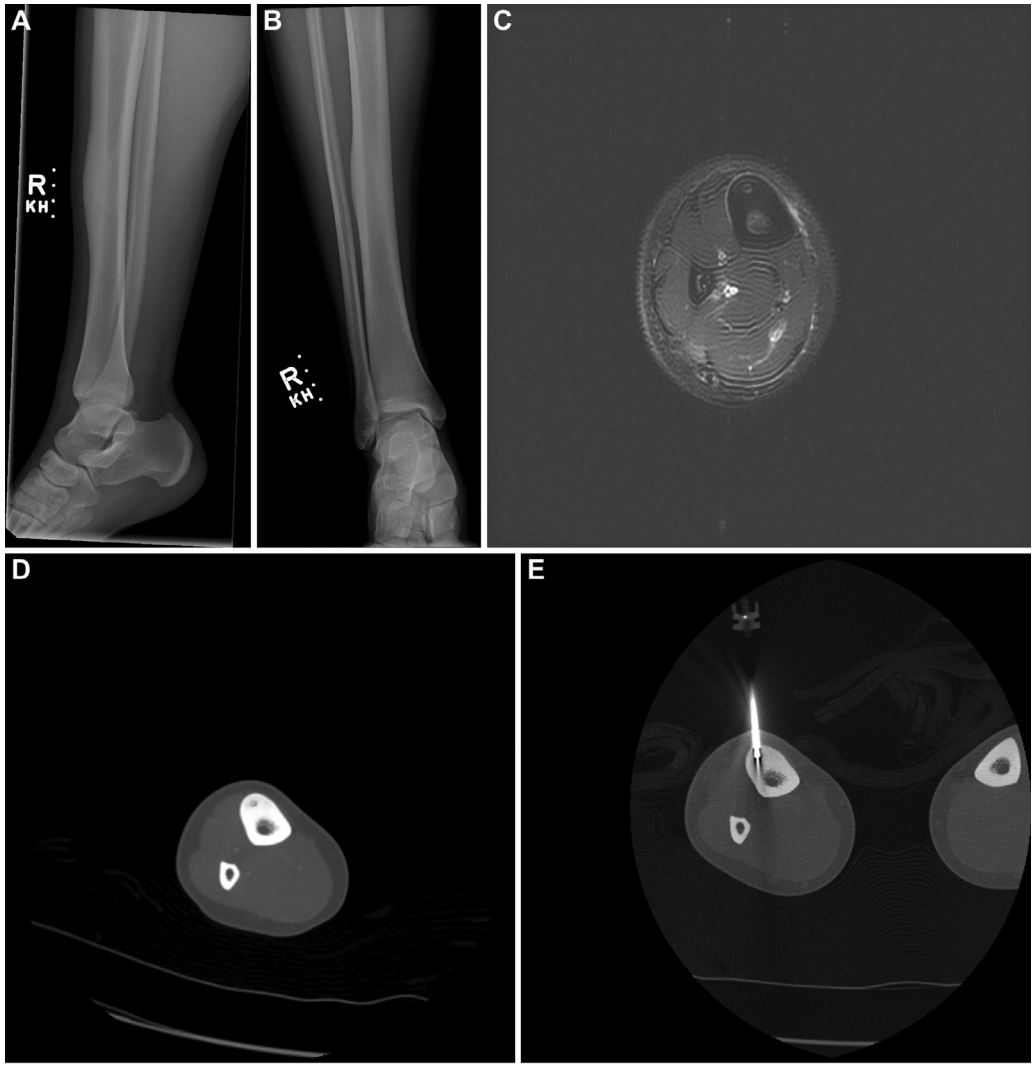

Fig.1 – Osteoid osteoma of the right tibia. (A and B) Anteroposterior (AP) and lateral radiographs demonstrate cortical thickening along the anterior mid tibial diaphysis. (B) Axial fat-saturated T2-weighted image shows thickening of the anterior tibial cortex with a small, cortically based, well-circumscribed intermediate signal lesion. There is significant cortical thickening with periosteal edema. (D) Axial CT shows a small, well-defined lucent lesion in the anterior midshaft of the right tibia surrounded by a thickened cortex, confirming the diagnosis of an osteoid osteoma. (E) Intraprocedural axial CT of radiofrequency ablation of the right tibial osteoid osteoma.

a case of metachronous osteoid osteomas occurring in separate bones which were successfully treated by percutaneous radiofrequency ablation (RFA).

Case report

An otherwise healthy 16-year-old female tennis player presented with progressive right leg pain which was worse in the evening, particularly after running. Physical exam revealed a palpable, tender mass over the anterior aspect of the tibia. Radiographs demonstrated pronounced cortical thickening of the distal tibial diaphysis (Fig. 1A and B). An MRI of the right tibia was ordered (Fig. 1C), because of concern for a stress fracture. The MRI showed periosteal reaction and a small cortically based lesion, suspicious for an osteoid osteoma, which was confirmed with a subsequent CT (Fig. 1D). The patient underwent RFA of the lesion (Fig. 1E) with good response and was able to return to athletic activities.

Approximately 4 months after undergoing RFA of her right tibia, the patient began to experience anterior right hip pain. Based upon her physical exam findings, the initial working diagnosis was flexor tendonitis of the right hip. Radiographs and an MRI of the right hip were performed for confirmation. While the radiographs showed no distinct explanation for the patient's symptoms (Fig. 2), the MRI showed edema within the anterior right acetabulum (Figs. 3A and 4A). This was felt to represent a stress fracture with edema extending to the adjacent iliopsoas muscle and tendon, thus explaining the patient's symptoms.

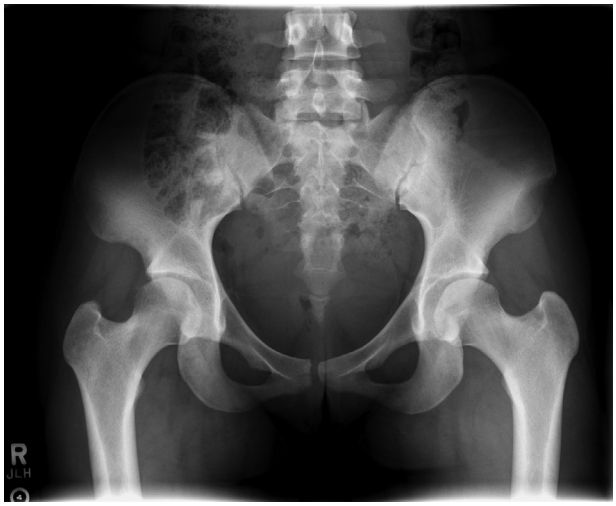

Fig. 2 – Pelvic radiograph obtained for right hip pain. An AP radiograph of the pelvis obtained to evaluate the patient's right hip pain shows no significant abnormality.

The patient followed weight-bearing precautions and participated in physical therapy. However, her pain continued to worsen, despite an interval MRI exam demonstrating decreasing marrow edema (Fig. 3B). Due to her ongoing symptoms, an MRI was performed 11 months after she first experienced symptoms at the right hip. This MRI revealed a circumscribed, peripherally T2 hyperintense, centrally T1/T2 hypointense lesion in the right anterior acetabulum, at the site of the prior marrow edema (Fig. 4A and B). As this MRI raised concern for an osteoid osteoma, a CT was suggested, which demonstrated a 1.1 cm lucent lesion with a central sclerotic focus and surrounding sclerosis in the right anterior acetabulum, consistent with an osteoid osteoma (Fig. 5A). The patient underwent RFA of the acetabular lesion soon after diagnosis (Fig. 5B). Intraprocedural biopsy confirmed the diagnosis of osteoid osteoma (Fig. 6).

The patient has recovered well from treatment. At 2 months follow-up postablation, she reports being pain-free and has returned to participating in competitive collegiate tennis.

Discussion

The occurrence of multiple osteoid osteomas is rare, first described by Scehawjowicz in 1970 [3]. When multiple osteoid osteomas occur in the same patient, it is usually within the same bone [8]. To our knowledge, our patient is only the seventh reported case in the medical literature of multiple osteoid osteomas occurring in 2 widely separate bones [7,8]. In contrast, we found 23 reported cases of multiple osteoid osteomas located within the same bone and 6 reported cases of osteoid osteomas in adjacent bones. We believe this distinction is important because the underlying etiology for these 2 patterns of multicentric presentation may be different. Some authors have proposed that monostotic multifocal lesions are due to an attempted healing response which divides 1 nidus into nidi. An alternative explanation postulates that the separate lesions may represent an evolving stage in the possible progression to osteoblastoma [7,9]. Obviously, neither of these 2 theories would explain the pathogenesis of polyostotic osteoid osteoma. To date, the precise etiology for both solitary osteoid osteoma and for polyostotic osteoid osteoma remains unknown [10].

The general indication for treatment of osteoid osteomas is persistent pain. Historically, treatment has involved surgical en bloc excision. Since its introduction for the treatment of osteoid osteoma in 1992, RFA has quickly become widely accepted treatment method [11,12]. Many now consider RFA as the treatment of choice for osteoid osteomas as it is a simple, minimally invasive procedure with clinical success rates

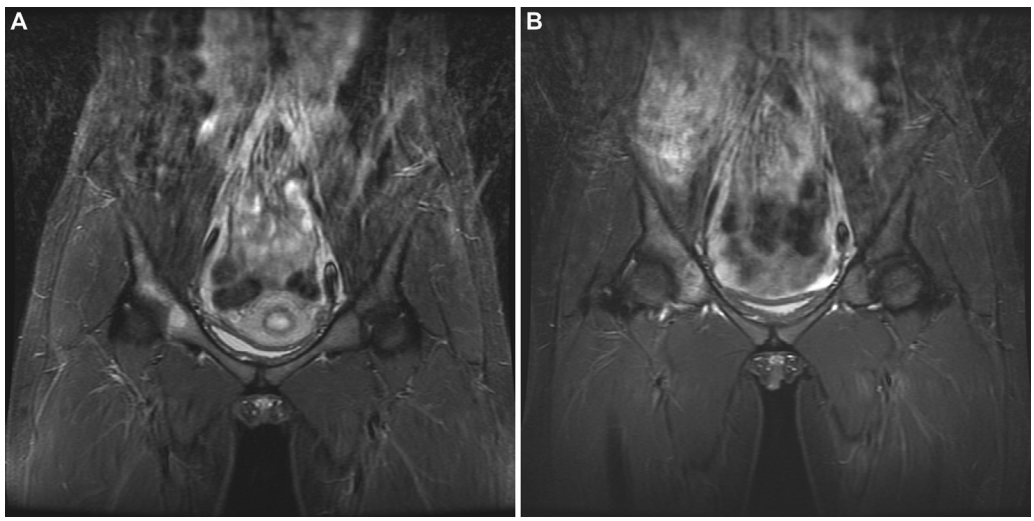

Fig. 3 – Coronal STIR images of the pelvis at initial presentation (A) and 6 months after presentation (B) demonstrate progressively decreasing marrow edema in the right acetabulum.

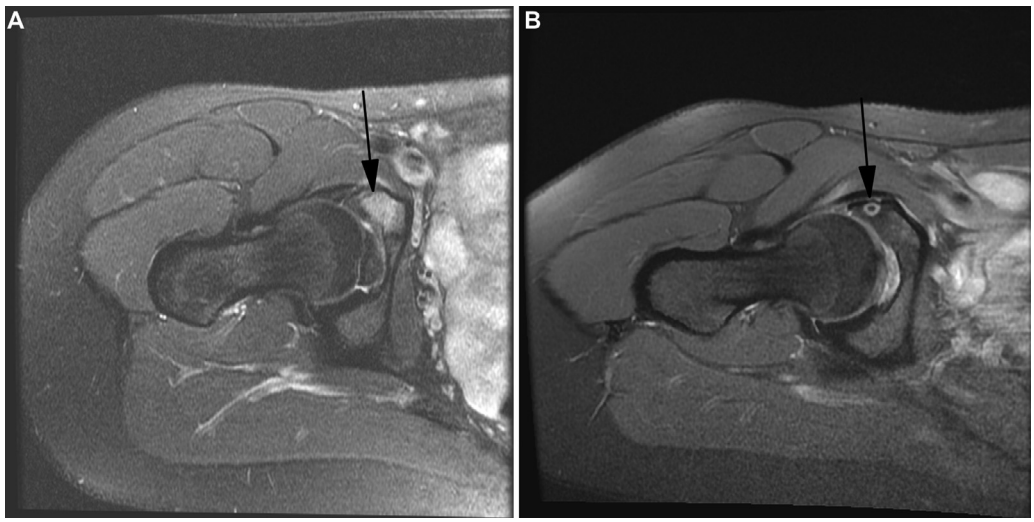

Fig. 4 – MRIs of the right hip at initial presentation and 11 months after initial presentation. (A) Axial fat-saturated proton density-weighted image at initial presentation demonstrates marrow edema in the right anterior acetabulum (arrow). (B) Axial fat-saturated proton density-weighted image at 11 months after presentation shows a discrete, peripherally hyperintense, centrally hypointense lesion in the region of the prior edema (arrow).

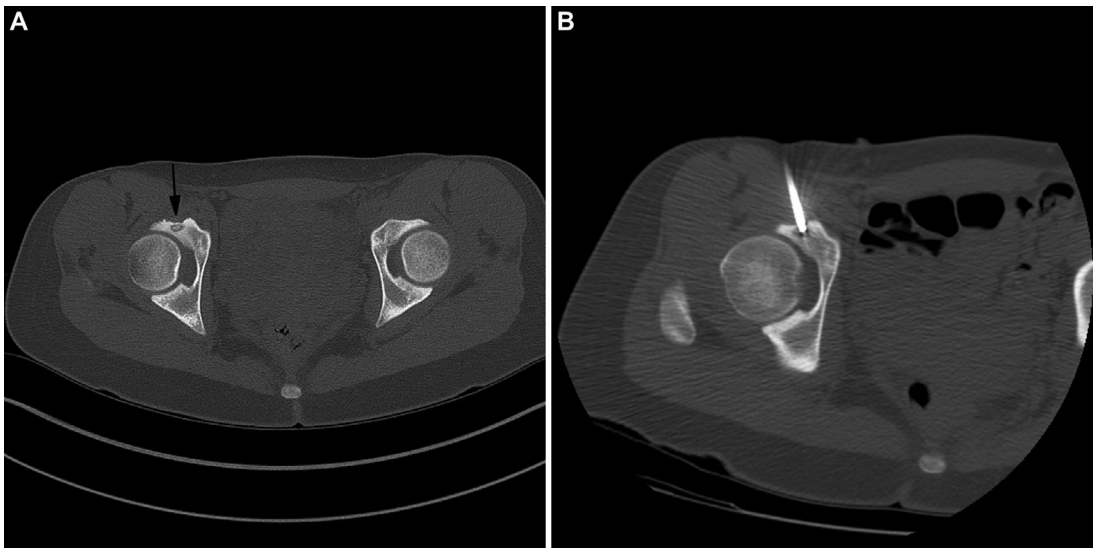

Fig. 5 – Axial CTs of the right acetabular osteoid osteoma. (A) Small lucent lesion with a central sclerotic focus and surrounding sclerosis in the right acetabulum, consistent with an osteoid osteoma. (B) Intraprocedural axial CT of subsequent radiofrequency ablation of the right acetabular osteoid osteoma.

of 89%-95% [12,13]. In our patient, both of her lesions were successfully treated with RFA.

Our case demonstrates the potential limitations of MRI in the diagnosis of osteoid osteoma. Our patient's diagnosis of her second osteoid osteoma was made 11 months after initially presenting with hip pain. Delay in diagnosis is not uncommon with periarticular osteoid osteomas as compared with extra-articular lesions [14,15]. The delay in diagnosis in our case was primarily due to obscuration of the tumor nidus by marrow edema on the initial MRIs. Multiple studies have demonstrated similar limitations of MRI compared to CT [4,16,17]. Small nidi can be difficult to detect on MRI due to the similar signal characteristics of the nidus to the adjacent cortex as well as the decreased spatial resolution of MRI. Although MRI is superior in depicting intramedullary and soft tissue changes, this may cause a misleading aggressive appearance of these benign tumors or obscure the nidus altogether. In contrast, the nidus is typically well-visualized on CT, but the intramedullary edema is undetectable. Although the spatial resolution of MRI is improving and may lead to improved detection of the central nidus in the future, CT remains the modality of choice for the diagnosis of osteoid osteoma [4]. In our patient, CT was able to demonstrate the small nidus in the acetabulum.

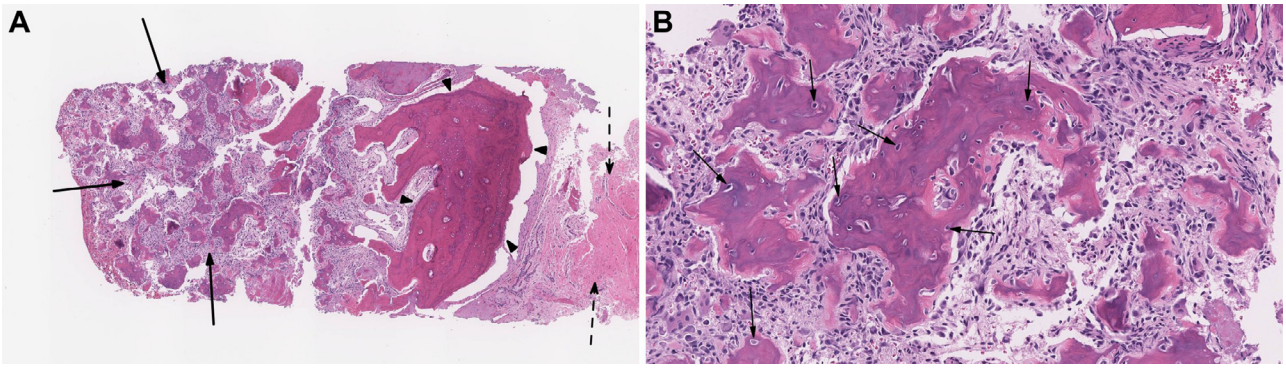

Fig. 6 – Microscopic photographs of the right periacetabular osteoid osteoma. (A) The characteristic central nidus of osteoid formation (solid arrows) with a rim of sclerotic bone (arrowheads) and adjacent fibrous tissue (dashed arrows) (H&E, 40×). (B) Higher magnification view of the nidus shows irregular osteoid with abundant associated osteoblasts (arrows) (H&E, 400×).

In conclusion, our case of multiple osteoid osteomas is unique for the following reasons: the polyostotic distribution of the tumors, the metachronous presentation, the periarticular location of the second lesion, and the use of RFA for treatment of both tumors. Our case suggests that multicentric osteoid osteoma should be included in the differential diagnosis in patients with a history of osteoid osteoma who present with musculoskeletal pain. Misdiagnosis or delayed diagnosis can be reduced by having a high index of suspicion and with appropriate use of CT imaging.

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