Multicentric Osteoid Osteoma Presenting a Diagnostic Dilemma

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We present a case of a relatively common benign tumor that was a diagnostic dilemma because of its atypical appearance in multiple imaging modalities. Our patient was a 22-year-old man who presented with complaint of three months of shin pain with running. The radiographically demonstrated sclerotic lesion in his right tibia initially was thought most likely to be a stress fracture, but on further evaluation, it had features that suggested a subacute osteomyelitis or Brodie’s abscess with focal sequestra. It was in fact, biopsy proven to be an osteoid osteoma with multiple, closely adjacent nidi. Its elongate, multicentric, “string of beads” arrangement as well as its intramedullary location is uncommon and it was larger than normally expected. Because of the unusual presentation and appearance of this lesion it caused a diagnostic dilemma. This lesion was evaluated with a full spectrum of modalities including radiographs, CT, MRI, Technetium 99m-MDP bone scan and Indium-111 white blood cell scan. After biopsy achieved a definite diagnosis, this tumor was successfully treated with radiofrequency ablation of all nidi in one session.

Osteoid osteoma is a relatively common benign bone tumor that accounts for approximately 12% of benign bone neoplasms [1]. It typically presents in the young adult or pediatric population. Although ages at presentation ranged from 19 months to 56 years in the Armed Forces Institute of Pathology study by Kransdorf et al, it is usually seen between the ages of 7 and 25 [1, 2]. A male predominance has also been demonstrated [1-3]. A characteristic history of night pain relieved with aspirin is seen in many, but is not universal [1]. Classically the tumor is found in the cortex of long bones with 50% or more arising in the femur or tibia, however it may arise in any bone and may be intramedullary or subperiosteal [2]. The nidus of the lesion is a round or oval...
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The radiolucent region usually less than one centimeter corresponding to the pathologic finding of interlacing trabeculae of bone and osteoid (uncalcified bone) with highly vascular, connective tissue stroma which has been demonstrated to contain neural elements and prostaglandin [2-6]. This radiolucent nidus may have a sclerotic center or central calcification and is surrounded by a variable zone of reactive/sclerotic bone [2,3,7].

The presentation and imaging features of these lesions, particularly the degree of reactive surrounding sclerosis varies with their bone of origin, their location with respect to the joint capsule; whether intra- or extracapsular. In 1966, Edeiken et al classified osteoid osteoma by the location of the nidus with respect to the cortex (cortical, cancellous or medullary, subperiosteal) [8]. In 1998, Kayser and Resnick et al further characterized the location of the nidus in tubular bones as subperiosteal, intracortical, endosteal, or medullary [9].

The location of the nidus with respect to the cortex, particularly if subperiosteal or intramedullary or within a joint can alter the radiographic appearance of the lesion and make the radiographic diagnosis of osteoid osteoma more challenging [2]. The medullary location of the lesion in our case is uncommon, shown in 2 of 38 cases in one study Kayser [9] and 6 of 67 in a second study by Klein [3].

In rare circumstances, a single osteoid

Figure 1. 27-year-old man with multicentric osteoid osteoma. AP (A) and Lateral (B) radiograph of right lower leg demonstrating non-specific sclerosis. When windowed optimally, the AP (C) and lateral (D) radiographs now suggest a lucent, lobulated intramedullary lesion with, serpiginous sclerotic margins surrounding the lucent nidi, which were better demonstrated on the following CT.
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osteoma may contain more than one nidus. Or multiple osteoid osteomas with discrete solitary nidi may be found in a single bone or neighboring bones [2]. A multiplicity of nidi in a multi-centric, “string of beads” arrangement has been described in fewer than 20 other English language case reports [10].

Case Report

A 22-year-old man initially presented with right shin pain of 3 months duration that was worse with running. The pain was localized to the medial aspect of the proximal third of the tibia. He stated that he felt a hard bump in the area and noted soft tissue swelling after running. He was initially worked up with conventional radiographs which demonstrated cortical thickening at the junction of the proximal and middle thirds of the tibia without evidence of lucency or fracture (Figs. 1A and 1B). A

Technetium 99m-MDP bone scan was recommended with suspicion for stress-related changes. In retrospect, the initial radiograph can be windowed in such a way to demonstrate serpiginous sclerosis about the two discrete lucent nidi within this lesion (Figs. 1C and 1D).

The bone scan showed an intense focus of increased radiopharmaceutical uptake in an ovoid distribution within the proximal right tibia which was centrally located in the lateral projection (Fig. 2). An extended pattern of mild, diffusely increased osseous uptake was observed in the surrounding bone. There was no significant cortical involvement. CT and MRI were recommended due to the concern for chronic stress fracture, fibrous dysplasia, or neoplasm.

seen on the CT was an elongated, lobulated appearing, intramedullary lucent lesion with linear ossified or calcified fragments in both the upper and lower aspect of the lucent channel suggestive of a sequestrum (Figs. 3A, 3B, 4A, and 4B). No soft tissue abnormality, masses or cortical breakthrough were demonstrated in conjunction with this lesion. The differential diagnosis was expanded based on these imaging findings to probable subacute osteomyelitis with Brodie’s abscess versus an atypical osteoid osteoma. An indium-111 white blood cell scan and correlation with CBC and ESR were recommended.

The MRI showed diffusely increased T2 signal within the proximal tibial diaphysis, with an elongated, slightly lobulated, geographically circumscribed area of increased T2 signal within the medullary aspect of the bone consistent with the nidi (Fig. 5A, 5B, and 5C). It measures approximately 9 mm in diameter and approximately 3.7 cm in total length along the long axis of the tibia. This area demonstrates intermediate to decreased signal on T1-weighted images (Fig 6A and 6B). On the axial T1 and T2 images, the medullary nidi can be seen to contain a central area of decreased T1 and T2 signal which may represent a central calcification or seques-
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trum (Fig 7A and 7B). No fracture, soft tissue mass, or expansion of the tibia was noted.

The appearance of the lesion on CT and MRI suggested chronic osteomyelitis, osteoblastoma, or an atypical osteoid osteoma. An orthopedic consult was placed as well an order for an indium-111 labeled white blood cell (WBC) scan. The WBC scan showed mild uptake in the proximal tibial diaphysis that was felt to be consistent with chronic intramedullary osteomyelitis (Fig. 8).

The orthopedic surgery consultant recommended a

Figure 3. Coronal CT reformations of right lower extremity showing a multi-lobulated, elongate lucent lesion in a medullary, diaphyseal location with a sclerotic margin. Note the linear ossific fragment in both the upper (A) and lower (B) nidus.

Figure 4. Axial CT through the medullary lesion in the proximal tibia demonstrating a well-circumscribed lucent nidus with small central ossification in both the upper (A) and lower (B) nidus.
biopsy, which was performed at an outside institution. Pathology confirmed a diagnosis of osteoid osteoma. The patient subsequently underwent radiofrequency ablation of the multiple nidi of the osteoid osteoma in a single session. His recovery was complicated with a wound infection, but more recent radiographs and CT have demonstrated cortical thickening and medullary sclerosis consistent with healing post radiofrequency ablation.

Figure 5. Sequential coronal T2-weighted MRI (A, B, C) showing increased T2 medullary signal about the lesion consistent with a large area of surrounding marrow edema/hyperemia.

Figure 6. Sequential Coronal T1-weighted MRI (A, B) demonstrating the elongate nature of the lesion with two of the adjacent nidi with surrounding low signal sclerosis and adjacent intermediate signal consistent with marrow edema.
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Figure 7. Axial T1 (A) and STIR (B) MRI demonstrate the medullary location of the nidus with a small central region of low signal consistent with calcification/ossification within the nidus.

Figure 8. Indium-111 labeled WBC scan demonstrated subtle increased cortical uptake in the right tibia and was felt to be consistent with chronic osteomyelitis but likely simply reflected the highly vascular connective tissue stroma within the nidus or adjacent marrow hyperemia.
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Discussion

In this case, our 22-year-old patient had 3 months of pain with running rather than the classic history of night pain relieved by aspirin. In our active military population, a stress fracture would be the most likely etiology. Further imaging demonstrated an intramedullary location with an elongate radiolucent lesion with two adjacent lobulations or nidi. His age and gender made osteoid osteoma a reasonable consideration, however the imaging findings of his lesion were not classic and there was diagnostic uncertainty. His lesion was in an unusual, medullary location and had an elongated multilobulated appearance with two adjacent nidi giving it a “string-of-beads” appearance which has been described [10, 11]. The longest dimension of the radiolucent cavity at 3.7 cm greatly exceeded the normal parameters ascribed for osteoid osteoma of less than 1.5 cm, but when allowing for several adjacent nidi, this would not exclude an elongate form of osteoid osteoma with two adjacent nidi. Reassuring findings were that the minor dimension of both nidi was under 1 cm and there were no aggressive features. Osteoblastoma is another benign bone neoplasm which may be considered in the differential diagnosis. It is less common than osteoid ostema accounting for perhaps 1% of primary bone tumors and 3% of benign bone tumors, [7] but is seen in the same age range (and also with a slight male predisposition). Sometimes osteoblastoma is radiographically and even pathologically indistinguishable from osteoid ostema [7]. In this tumor, the sclerotic/calcific material in the nidi was also elongate causing some overlap in appearance with a sequestrum of a subacute or chronic osteomyelitis or Brodie’s abscess. The vascular matrix within the nidus of the osteoid ostema or perhaps the adjacent hyperemic marrow demonstrated mild uptake on an Indium labeled WBC scan which was interpreted as evidence of an infectious etiology. The patient’s laboratory values for CBC and ESR were normal. In spite of diagnostic uncertainty due to the atypical appearance of this lesion on multiple imaging modalities, his case was properly managed with biopsy, resulting in the diagnosis of multicentric osteoid ostema. This benign tumor was then treated with radiofrequency ablation, addressing both of the nidi within a single session. His course was complicated with a post radiofrequency ablation wound infection, which resolved with antibiotics, and he has since done well.

It is hoped that this case will increase the awareness of the potential for unusual radiographic presentation of the relatively common, benign bone tumor, osteoid ostema. In addition to its more classic cortical location, osteoid ostema may present in a subperiosteal or medullary location as shown in this case [9]. Unusual imaging findings may also be seen if an osteoid ostema appears within a joint capsule, although that was not seen in this case. Additionally, although rare, osteoid ostema may have multiple nidi giving it a “string-of-beads” appearance as demonstrated in this case in multiple imaging modalities. Consider an atypical presentation of this relatively common benign bone tumor in your differential for well circumscribed lucent bone lesions with sclerosis or lesions with sequestra as there may be some overlap in appearance with Brodie’s abscess.

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