Fracture-Site Osteoid Osteoma in a 26-Year-Old Man

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Osteoid osteoma is a benign osteoblastic lesion of bone. Osteoid osteomas make up approximately 11% of all biopsy-analyzed primary bone tumors.[1] After Bergstrand[2] and then Jaffe[3] first described this tumor, it was more frequently reported in various parts of the human skeleton. Most often, the tumor occurs in the diaphyseal area of the long bones, particularly the femur and the tibia, but there are many reports of metaphyseal and epiphyseal involvement,[4] as well as occurrence in almost every bone in the body. Diagnosing osteoid osteoma can be a significant challenge. This tumor has occurred in unusual clinical backgrounds, which can make diagnosis even more difficult.

In this article, we report a case of osteoid osteoma within a tibial fracture callus, presenting with persistent pain after union. The patient provided written informed consent for print and electronic publication of this case report.

Case Report

A 26-year-old man was referred to our university hospital with the chief concern of a 4-year history of pain in the mid-diaphyseal part of the right leg. The pain had started 4 years after open reduction and internal fixation, with dynamic compression plate, of a closed tibiofibular fracture. The fracture was sustained in a motorcycle crash. The pain most often occurred during night sleep and it woke the patient. He was taking a daily dose of nonprescription ibuprofen 200 mg, usually before bedtime, and reported having developed an addiction to oral opium for the pain. Pain symptoms increased during the month before referral. The patient was pain-free for 4 years after fracture fixation. The plate was removed because of the significant pain, after radiographic studies established union. The only other significant medical histories were smoking and opium addiction.

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Am J Orthop. 2012;41(4):E57-E60.

On physical examination, a previous linear longitudinal anterolateral scar of surgery was noted in the mid-diaphyseal area of the tibia. No other skin change was apparent. Range of motion of knee and ankle joints was normal.

Laboratory studies included complete blood cell count, erythrocyte sedimentation rate and C-reactive protein (CRP) level. Erythrocyte sedimentation rate and CRP level were normal.

Radiographic study of the area of reported pain revealed circumferential cortical thickening in the site of the previous fracture, along with a suspicious lucency within the lateral cortex (Figure 1). Three-phase technetium-99m bone scan showed increased tracer uptake in the suspected area (Figure 2). Computed tomography (CT) revealed an area of increased density within the low-density area surrounded by dense cortical bone (Figure 3). The pattern raised the suspicion for osteoid osteoma with a small nidus or a sequestrum associated with osteomyelitis.

Given the patient's medical history and imaging studies, an excisional biopsy was performed. The previous incision site was used to expose the bulging cortex of

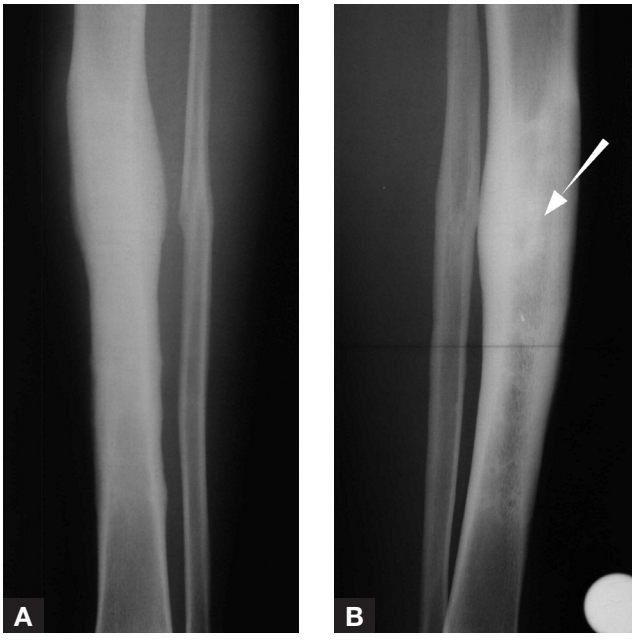

Figure 1. Anteroposterior (A) and lateral (B) radiographs of right leg show complete tibial fracture-site healing with suspicious lucency visible only on lateral view (arrow).

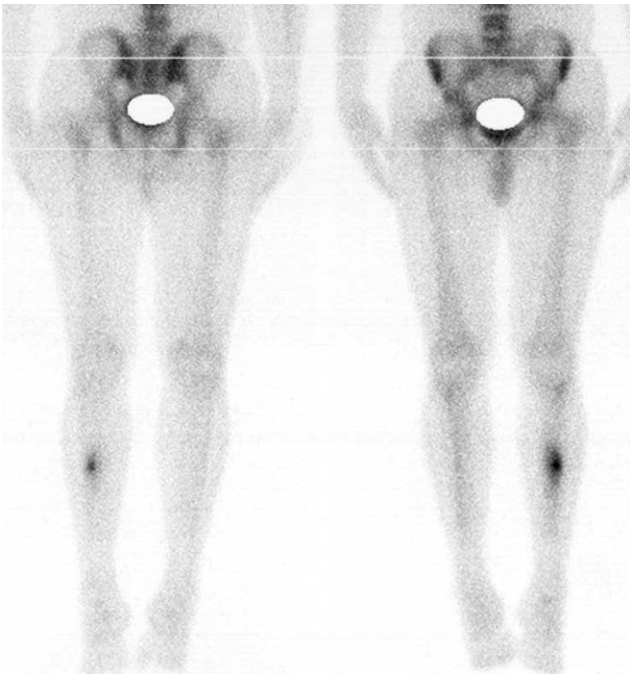

Figure 2. Technetium-99m whole-body bone scan shows increased tracer uptake in lateral aspect of tibial midshaft.

the tibial diaphysis and the cortex was burred down until a suspected red nidus was encountered. The nidus was excised and the specimen was sent for pathologic and microbiological study. The wound was irrigated and closed in routine fashion.

The pain relief after surgery was dramatic. The pathologic diagnosis was osteoid osteoma and the cultures were negative (Figure 4). At 2-year follow-up, the patient was still asymptomatic and radiographic study findings were insignificant.

Discussion

Osteoid osteomas comprise approximately 11% of all biopsy-analyzed benign primary bone tumors.[1] They usually occur during the second or third decade of life and are more prevalent in males. The most common presenting symptom is pain. Swelling, joint effusion, and limited range of motion can also be observed, particularly in para-articular and intra-articular lesions. In the hand, an osteoid osteoma can present with monoarticular arthritis, clubbing, or macrodactyly in the absence of sclerosis or

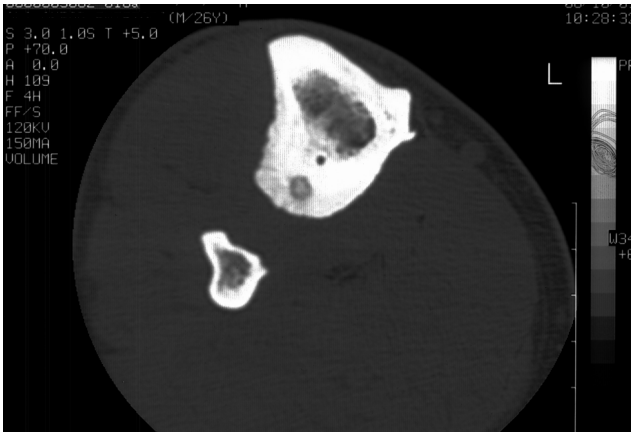

Figure 3. Computed tomography with axial cut through tibial lesion shows area of decreased density with surrounding sclerosis of fracture callus and relatively dense area within lesion in lateral cortex.

lytic lesion.[5,6] Osteoid osteoma can be found in the cortex, the medullary canal, or the periosteum, or it can be intra-articular. Many osteoid osteomas have been found with atypical presentations and in unusual locations.

The pathogenesis and nature of osteoid osteoma are not well understood. Whether osteoid osteoma is a true neoplasm, a reactive lesion in response to trauma, inflammation, or infection, or an unusual healing or vascularization process is a matter of debate.[7,8]

In some reports, the natural history of osteoid osteoma seems to be spontaneous remission,[3,7,9] though most of these reports omit a pathologic diagnosis. However, most patients require surgical ablation because they cannot tolerate the pain.

Information on the role of traumatic events in the pathophysiology of osteoid osteoma is open to doubt. Primary reports do not mention any etiologic relationship between trauma and formation of osteoid osteoma.[10] More recently, however, some reports have documented osteoid osteoma occurring after traumatic events or fractures.[11-15]

Cases of fracture-site osteoid osteoma with pathologic confirmation are summarized in the Table and described here. In a pioneering report, Garcia and colleagues[12] presented the case of a 24-year-old man with thigh pain characteristic of osteoid osteoma 4 years after femoral shaft fracture and 2 years after intramed-

Table. Summary of Reported Cases of Fracture-Site Osteoid Osteoma

Reference	Age, y	Sex	Site	Postfracture Interval, y	Additional Information
Garcia et al[12] (1981)	24	M	Femur	4	—
Adil et al[11] (1996)	22	M	Distal radius	6	No tissue culture provided
Soon et al[15] (1999)	24	M	Tibia	5	—
Grenard et al[13] (2001)	18	F	Femur	2	—
Schulze et al[14] (2001)	14	F	Distal tibia	3	—
Our patient (2012)	26	M	Tibia	8	—

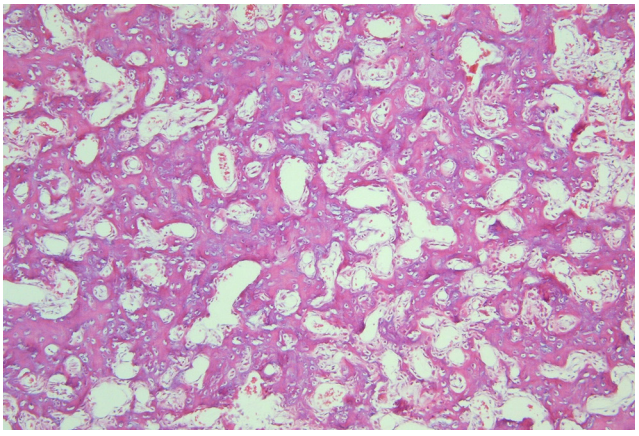

Figure 4. Abundant vascular structures and immature bone spicules lined with osteoblasts and osteoclasts (hematoxylin-eosin stain, original magnification ×400). Pathologic diagnosis was osteoid osteoma.

ullary rod removal. The diagnosis was suspected with radiologic findings and confirmed with pathologic study after en bloc resection led to symptom resolution.

Baron and colleagues[16] reported finding osteoid osteoma in the tibias and femurs of 2 young adults who presented with pain. Both patients had a previous surgical procedure at the site. In one case, the lesion was found in the track of the previous screw fixation. The diagnosis was apparently delayed, suspected with imaging studies, and confirmed with surgical resection and pathologic study. The authors used several criteria to define post-traumatic osteoid osteoma: initial trauma, silent period, suggestive pain, discovery of osteoid osteoma at trauma site, and recovery after surgical treatment. In their review of the literature, they identified 13 more patients who met these criteria, and they discussed the role of traumatic events in the evolution of osteoid osteoma.

Adil and colleagues[11] subsequently reported the case of a 22-year-old man with wrist pain and a 6-year history of distal radial fracture with exuberant callus formation. The fracture had been treated with rod insertion. En bloc resection showed osteoid osteoma in pathologic study. The authors speculated that invagination of the periosteum during trauma, fracture, reduction, or pinning may be a predisposing factor for development of osteoid osteoma. Unfortunately, they did not perform a microbiological study on the specimen, and their report does not mention blood leukocyte count or CRP level. Lund[17] found that osteoid osteoma and osteomyelitis are similar with respect to histopathologic and radiologic features and they reported 2 cases of osteoid osteoma associated with osteomyelitis.

Grenard and colleagues[13] found an osteoid osteoma in the site of femoral fracture union in an 18-year-old girl. The symptom—pain, mostly nocturnal, with response to aspirin—started after the plate osteosynthesis was removed, about 2 years after injury. During that postoperative period, the wound began giving off a serosanguinous discharge, which resolved with local wound care. Computed tomography and bone scan raised the suspicion of osteoid osteoma. Leukocyte-labeled bone scan was negative. The authors emphasized the importance of considering low-grade bone infection and osteomyelitis in the diagnosis of osteoid osteoma.

Soon and colleagues[15] reported osteoid osteoma in a previous site of tibial stress fracture in a 24-year-old man. The stress fracture was diagnosed by radiography and bone scan. Pain persisted for 5 years before the patient underwent surgery, received a pathologic diagnosis of osteoid osteoma, and experienced symptom resolution.

Schulze and colleagues[14] found osteoid osteoma in a previous site of distal tibial open fracture in a 14-year-old girl. The patient was referred with a report of pain 3 years after injury and hardware removal. Given her medical history and radiologic studies, osteomyelitis was included in the differential diagnosis. Blood leukocyte count and CRP level were normal. Histopathologic study and specimen cultures did not show evidence of infection. Pain resolved after lesion resection.

In our patient, the infectious pathology was ruled out. Given the data in the literature and the reports described here, the association between traumatic events (eg, fracture) and osteoid osteoma remains enigmatic. In our patient's case, as in other cases, there was no evidence of a lesion on radiographs obtained immediately after fracture or during healing/follow-up, and the pain started long after the injury. The diagnosis is typically delayed because there is a low index of suspicion for the tumor. The periosteal invagination hypothesis of Adil and colleagues[11] has not been substantiated histologically in reported cases, including ours. Spjut[18] described a vascularization theory of development of osteoid osteoma in which abnormal bone tissue replaces normal tissue in a background of impaired vascularization, and leads to nidus formation. Uehlinger[19] proposed 3 phases in the development and differentiation of multicentric osteoid osteomas: (1) development of a cavity, (2) filling of the cavity by osteogenic mesenchyma with differentiation of an osteoid network and peripheral progressive osteoclasia, and (3) growth stop at a diameter of 8 to 10 mm with continuation of inner alterations. Although both periosteal invagination (Adil and colleagues[11]) and impaired vascularization (Spjut[18]) can lead to cavity formation (Uehlinger's[19] step 1), no substantiated study has confirmed this.[14] Our patient's osteoid osteoma was found in the lateral aspect of the tibial fracture site, where the internal fixation device was introduced.

The unclear pathophysiology of the tumor, the wide range of anatomical areas involved, the diversity of clinical backgrounds, and the various forms of clinical presentation all contribute to missed diagnoses of osteoid osteoma. Significant delays (11-36 months) between symptom onset and diagnosis have been reported.[20]

For patients who have had unexplained pain for a

long time after bone fracture treatment and particularly in cases involving internal fixation, the fracture site must be carefully scrutinized to evaluate for the presence of osteoid osteoma as an unusual "complication." This tumor should be considered as a differential diagnosis.

Our review of the literature regarding posttraumatic osteoid osteoma strongly suggests an association between traumatic events and pathophysiology of this tumor. Further research is warranted to elucidate pathophysiological aspects of this tumor.

Authors' Disclosure Statement

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

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