Case Report

Osteoid osteoma is a benign primary bone tumor containing a small region of abnormal osteoid with osteoblasts and variable surrounding reactive bone. Osteoid osteoma accounts for approximately 11% of all benign tumors. The majority of osteoid osteomas are monofocal with a single nidus. The orthopedic and radiology literature is scattered with several case reports of multicentric osteoid osteoma lesions. This article reports a case of an osteoid osteoma with a single nidus that progressed to a multicentric osteoid osteoma.

CASE REPORT

A 19-year-old man was referred with a 2-year history of right thigh and knee pain. He reported no history of trauma to the right thigh and had no fever, chills, night sweats, weight loss, or other constitutional symptoms. The pain was most bothersome during the night and occasionally in the early hours of the morning. An examination was within normal limits, and a presumptive diagnosis of osteoid...
When the patient presented 2 years ago at the outside institution, a magnetic resonance image (MRI) was ordered as the sole diagnostic study. The MRI showed a solitary lesion in the right medial mid-diaphyseal femur with a single nidus (Figure 1A). Treatment with nonsteroidal anti-inflammatory (NSAIDS) medications was started with good initial relief of pain.

Several months later, despite continued NSAID usage, the patient noted recurrence of right thigh and knee pain. Approximately 1 year after the initial study, a second MRI showed a second nidus located close to the initial lesion (Figure 1B). The NSAID treatment was continued, and he subsequently was referred to our medical center.

On clinical examination, the patient ambulated with a normal reciprocal gait pattern. Examination of the right lower extremity showed atrophy of the quadriceps musculature. The right mid-thigh circumference was 40 cm compared to 44.5 cm on the left. There was mild tenderness on palpation of the right medial mid-thigh. No soft-tissue mass was appreciated.

Motor strength of the hip flexors and knee extensors was 4/5. No limb-length discrepancy, inguinal lymphadenopathy, or knee effusion was present, and the patient had full, painless range of motion of his right hip and knee.

Radiographs of the right femur showed increased sclerosis of the medial mid-diaphysis of the right femur with two distinct nidi (Figure 2). Each lesion was approximately 1 cm in diameter. A computed tomography (CT) scan of the right femur also demonstrated the two separate nidi with surrounding sclerosis (Figure 3). A chest radiograph was normal. A technetium-phosphate bone scan showed two separate small focal regions of increased radio-labelled tracer uptake in the right mid-femur.

The radiographic differential diagnosis included osteoid osteoma, osteoblastoma, infection, and eosinophilic granuloma. Because the initial nonoperative treatment consisting of NSAIDs no longer alleviated the patient’s discomfort and he did not desire fur-
ther nonoperative therapy, a decision was made to proceed with open biopsy of the lesions, with treatment to be determined by the results of the histopathologic examination of the lesions.

The location of the lesions was confirmed with fluoroscopic assistance. A 6-cm longitudinal anteromedial incision was made over the two lesions. Dissection was performed through the thigh musculature and splitting of the vastus medialis oblique. Using image intensification, a 4.5-mm drill bit was used to drill the cortex. The lesions were curetted and burned under fluoroscopy to confirm complete removal of the lesions. No communication between the two lesions was noted during surgery.

Separate frozen and permanent specimens were sent to pathology. All specimens showed osteoid osteoma and reactive bone. The final pathology was consistent with multifocal osteoid osteoma (Figure 4). Osteoset (Wright Medical Inc, Arlington, Tenn) pellets were packed into each lesion, and Grafton (Synthes, Pa) demineralized bone matrix putty was placed on the Osteoset pellets and intact bony cortex.

Postoperatively, the patient was placed in a knee immobilizer and started initially with toe-touch weight bearing. His weight-bearing level increased to full weight bearing over approximately 6 weeks. One year after the index procedure, the patient remained without any complications or recurrences.

**DISCUSSION**

Osteoid osteoma is a benign primary bone lesion that usually affects individuals during the first three decades of life, with an increased occurrence in males (2:1). Pain caused by the lesion is classically described as night pain that is relieved by salicylates and NSAIDS. This relief possibly is the result of diurnal production of prostaglandins by the tumor. Radiographically, osteoid osteoma typically is described as a focal sclerotic bony lesion with a nidus <1 cm in diameter.

Spontaneous resolution over a variable amount of time has been proposed to be the natural history of the lesion. If the symptoms are not responsive to medication or the medication is not tolerated, the lesion can be treated surgically. Surgical treatment usually consists of curettage or en bloc resection, and in some centers, additional adjuvant agents such as phenol have been used.

Over the past decade, radiofrequency ablation with CT guidance has gained wide acceptance as a minimally invasive means of treatment. In our case, given the two distinct lesions and potential of other diagnosis, open surgical biopsy and curettage of the lesions was deemed prudent.

Approximately a dozen cases of multicentric osteoid osteoma have been reported in the literature. This is believed to be an underestimation because the multicentricity of the lesion may be missed.

Previous cases of multicentric osteoid osteoma note occurrences in unusual locations such as the ethmoid and pubis bone. Schai et al reported a case of a discrete synchronous multifocal osteoid osteoma of the humerus with two different foci. One focus was located in the cancellous region of the greater tuberosity and the second focus was located at the proximal humeral diaphysis.

Gonzalez et al reported a case in which radiographs of a 16-year-old boy showed a diffuse increase in bone density with cortical thickening of the distal right tibia. Diagnostic radiographic studies and operative specimens were consistent with a multifocal osteoid osteoma.

Gonzalez et al noted the pathologic significance of multifocal osteoid osteoma is unknown, and some authors suggest it may be an intermediate stage between osteoid osteoma and osteoblastoma. Schajowicz et al classified multifocal osteoid osteoma as multifocal sclerosing osteoblastoma.

Our case is unique in that the initial MRI demonstrated only a single nidus, which over the course of approximately 1 year progressed to a multicentric nidus. This progression has not been documented previously.

We believe this reported multicentric osteoid osteoma likely began as a solitary focus. It is possible a second distinct osteoid osteoma arose or the initial nidus divided into two separate foci. This may represent an intermediate stage in the progression of an osteoid osteoma to an osteoblastoma.

Alternatively, it could represent an incomplete attempt at healing that resulted in the walling off of the first nidus and the subsequent formation of two distinct nidi. Finally, because the etiology of osteoid osteoma remains unknown, it is possible a multicentric osteoid osteoma may be a manifestation representing a favorable local host response that leads to the development of an osteoid osteoma in a specific area of bone.

Given the rarity of multicentric osteoid osteoma, these questions will remain unanswered. However, because of the potential of a single nidus becoming multicentric, periodic imaging of osteoid osteoma appears worthwhile if the lesion is being treated conservatively or if it remains recalcitrant to medical management.

**REFERENCES**


