



# Metachronous Osteoid Osteoma of the Mid-Diaphysis of the Fibula and Distal Humerus: A Case Report

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## Abstract

### Keywords

- ▶ metachronous
- ▶ osteoid osteoma
- ▶ fibula
- ▶ humerus

Osteoid osteomas are mostly solitary. We report a case of metachronous osteoid osteoma of mid-diaphysis of fibula followed by distal humeral osteoid osteoma after a 7-year interval.

## Introduction

Osteoid osteomas are almost always solitary, with multiple tumors occurring in the same patient in less than 1% of reported cases.<sup>1,2</sup> Where multi focal tumors are encountered, they are typically found in the same bone<sup>1</sup> with up to 60% of cases being identified in the proximal femur and mid-diaphysis of the tibia.<sup>3</sup> The second commonest site is in the spine where up to 20% of cases occur, particularly in the posterior elements.<sup>4</sup> We report a rare case of metachronous osteoid osteoma of the mid-diaphysis of the right fibula followed by right distal humerus osteoid osteoma after a 7-year disease-free interval.

## Case Report

A 7-year-old boy presented with a 4-week history of pain and swelling of the right leg. He had a history of mucoepidermoid carcinoma of the parotid gland, which was treated by surgical resection without any complication. There was no other significant past medical history. A radiograph of the right leg

showed a lytic lesion of the right fibula diaphysis with associated periosteal reaction (▶ **Fig. 1**). A magnetic resonance imaging (MRI) of the right leg showed marked cortical thickening with osseous and soft tissue edema consistent with an osteoid osteoma of the fibula (▶ **Fig. 2**). The patient subsequently underwent Jamshidi biopsy and curettage of the lesion with the histopathological results of the biopsy showing fragments of a typical osteoid osteoma nidus with irregular calcified bony lamellae, surrounded by prominent osteoblasts and with loose vascularized surrounding stroma containing small osteoclasts (▶ **Fig. 3**). Symptoms resolved after the procedure and he was ambulating without any discomfort or pain.

The patient remained asymptomatic until he returned 7 years later with right elbow swelling, pain, and reduced range of movements for a period of 2 to 3 months. On examination, his elbow joint was grossly swollen, with no erythema. His elbow extension was limited lacking the final 35 degrees of extension. Pronation and supination movements were well preserved. A radiograph of the elbow joint demonstrated a large effusion with a sclerotic lesion in the

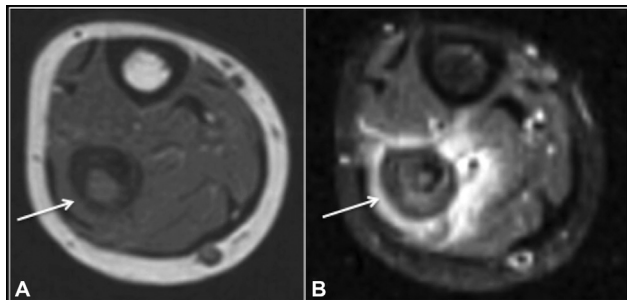
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**Fig. 1** Right leg anteroposterior radiograph showing osteoid osteoma (arrow) with marked cortical thickening.

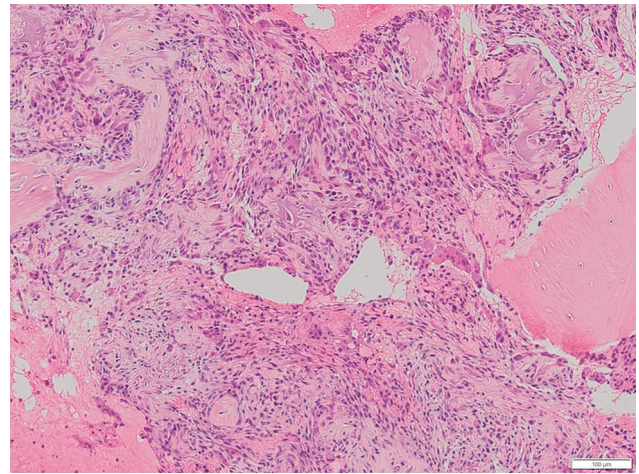


**Fig. 2** Axial T1 (A) and short tau inversion recovery (B) showing the osteoid osteoma (arrow) of the fibula with marked cortical thickening and osseous/soft tissue edema.

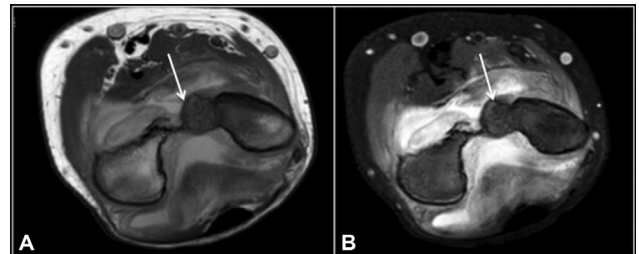
coronoid fossa of the distal humerus in keeping with an osteoid osteoma. An MRI was performed that showed a 1.3 × 1.4 cm lesion in the distal humerus at the level of coronoid fossa. There was a large joint effusion and surrounding bone marrow edema (► **Fig. 4**). The findings were discussed at a multidisciplinary meeting and a computed tomography (CT)-guided biopsy was organized, which confirmed the initial impression of an osteoid osteoma (► **Fig. 5**). Subsequently, a radio frequency ablation was performed and the patient recovered well following the procedure with complete resolution of pain.

## Discussion and Conclusion

Osteoid osteoma is an uncommon neoplasm of the bone accounting for ~10% of benign bone tumors.<sup>5</sup> It was first described by Jaffe in 1935.<sup>6</sup> Patients classically present with



**Fig. 3** Histological examination shows fragments of a nidus of an osteoid osteoma consisting of irregular trabeculae of woven bone lined by plump osteoblasts and the intervening stroma consisting of loose fibrovascular stroma.

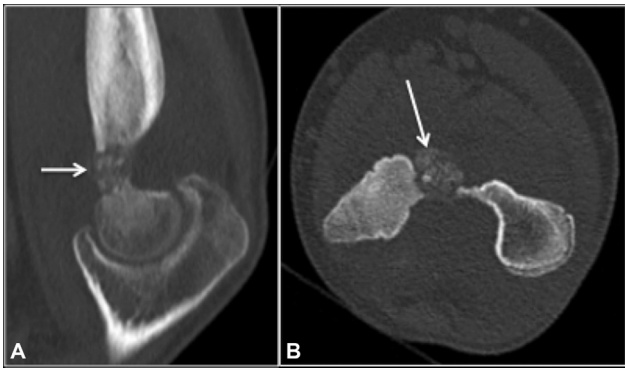


**Fig. 4** Axial T2 (A) and Proton Density Fat Saturated (PDFS) Sequences (B) showing osteoid osteoma in the distal humerus (arrow) with and associated large joint effusion and synovitis.

persistent dull achy pain worse during the night relieved with aspirin or nonsteroidal anti-inflammatory drugs. It is most commonly seen in adolescents and young adults in a 2:1 male to female ratio.<sup>7</sup> The treatment of symptomatic osteoid osteoma usually involves radio frequency ablation or curettage.

The classic radiological appearances of osteoid osteoma are of a radiolucent oval central focus with a size of smaller than 1 cm with surrounding reactive sclerosis.<sup>8</sup> CT may show a well-defined nidus of low attenuation, but higher attenuation may be seen in cases where the nidus is mineralized.<sup>8</sup> MRI demonstrates the nidus with marked perilesional osseous and soft tissue edema and surrounding sclerosis.<sup>9</sup> A joint effusion and synovitis may also be seen.

The occurrence of multiple osteoid osteomas is rare. The majority of cases were found to be localized to the same region/bone.<sup>10</sup> Schai et al reported a case of multifocal osteoid osteoma with one focus in the cancellous region of the greater tuberosity of the humerus and a second cortical focus at the proximal humerus diaphysis.<sup>2</sup> In very rare cases, multiple synchronous tumors may be found in adjacent bones and to a lesser extent in remote bones.<sup>11</sup> Beck et al reported a metachronous osteoid osteoma of the tibia and the T7 vertebral body, while Giuseppe et al described a case



**Fig. 5** CT Sagittal (a) and axial (b) showing osteoid osteoma in the distal humerus (arrow).

of osteoid osteoma of the right distal femur with a history of previous osteoid osteoma in the left distal femur.<sup>11,12</sup> In our patient, the lesions were found to be anatomically distant and were separated by a 7-year interval. It is possible that this occurrence represents nothing more than a mere coincidence; however, it is also possible that a metachronous presentation of these tumors reflects a genetic predisposition.

Our case indicates that a second osteoid osteoma may occur in a different bone in the same patient even after many years. Therefore, in the case of a patient with previous history of OO presenting with typical signs and symptoms of osteoid osteoma such as deep and persistent pain at a different site, worsening at night, and relieved by salicylates, it is important to include a metachronous osteoid osteoma as part of the differential diagnosis.

#### Conflict of Interest

No conflicts of interest.

No financial disclosures.

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