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.1,2 Where multi focal tumors are encountered, they are typically found in the same bone1 with up to 60% of cases being identified in the proximal femur and mid-diaphysis of the tibia.3 The second commonest site is in the spine where up to 20% of cases occur, particularly in the posterior elements.4 We report a rare case of metachronous osteoid osteoma of the mid-diaphysis of the right fibula followed by right distal humeral 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 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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coronoid fossa of the distal humerus in keeping with an osteoid osteoma. An MRI was performed that showed a 1.3 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.\(^5\) It was first described by Jaffe in 1935.\(^6\) Patients classically present with 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.\(^7\) 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.\(^8\) CT may show a well-defined nidus of low attenuation, but higher attenuation may be seen in cases where the nidus is mineralized.\(^8\) MRI demonstrates the nidus with marked perilesional osseous and soft tissue edema and surrounding sclerosis.\(^9\) 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.\(^10\) 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.\(^2\) In very rare cases, multiple synchronous tumors may be found in adjacent bones and to a lesser extent in remote bones.\(^11\) Beck et al reported a metachronous osteoid osteoma of the tibia and the T7 vertebral body, while Giuseppe et al described a case...
of osteoid osteoma of the right distal femur with a history of previous osteoid osteoma in the left distal femur.\textsuperscript{11,12} 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.

References