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CASE REPORT

Osteoid osteoma transformation into osteoblastoma: Fact or fiction?

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KEYWORDS
Osteoid osteoma;
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Child;
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Summary
Background: Osteoid osteoma and osteoblastoma are rare, benign, bone-forming tumours. The clinical presentation, imaging study findings, and course indicate clearly that these two tumours are distinct entities.
Clinical reports: We report two cases suggesting transformation of osteoid osteoma into osteoblastoma and therefore inviting a discussion of the links between these two tumours. An 11-year-old girl with a small metaphyseal lesion of the proximal tibia was given a diagnosis of osteoid osteoma. Over the next few weeks, worsening pain and marked tumour growth prompted a biopsy, which was consistent with an aggressive osteoblastoma. A review of the case suggested primary osteoblastoma at the earliest stage of development. In a 14-year-old boy, en-bloc excision was performed to remove a 1 cm defect located within the femoral shaft cortex and typical for osteoid osteoma. An asymptomatic recurrence measuring 20 mm along the long axis was removed 18 months later. Reassessment of the histological slides indicated recurrence of an incompletely excised osteoid osteoma.
Discussion: The histological similarities between osteoid osteoma and osteoblastoma, together with the lesion size criterion, may result in confusion. Collaboration between the clinician and pathologist is crucial and should take the tempo of evolution into account.
Conclusion: The histopathological differences between these two tumour types deserve to be emphasized. The data reported here challenge the concept that osteoid osteoma can transform into osteoblastoma. These two tumours are distinct entities that should no longer be differentiated based on size, as was long done in the past.

Introduction

Osteoid osteoma and osteoblastoma are rare, benign, bone-forming tumours that account for about 12% and 3%, respectively, of all benign bone tumours [1,2]. In the past,

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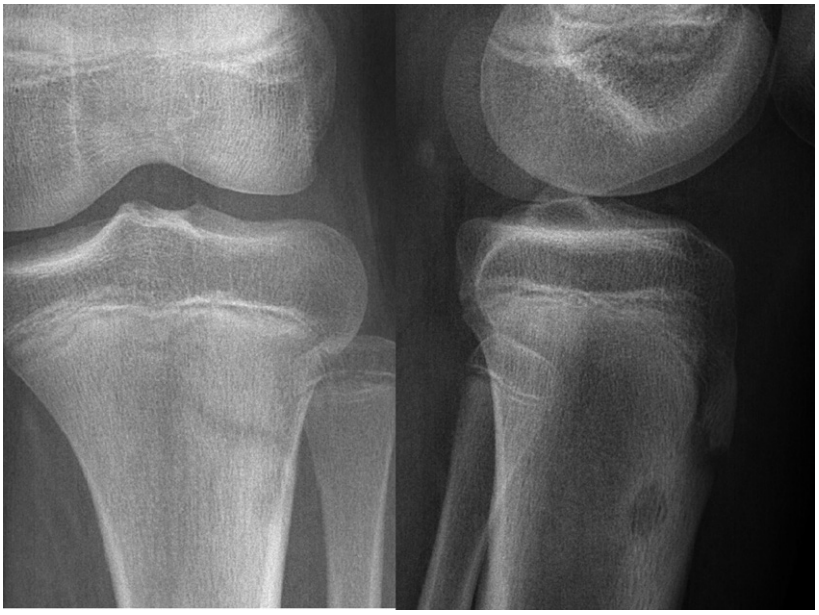

Figure 1 Case #1: radiograph of the knee showing a 1 cm defect in the lateral cortex of the proximal tibia, with minimal rim sclerosis.

these two tumours were classified as variants of a single tumour type, based on a number of shared histological features [3—5]. Lesion size was long used as a discriminating criterion, with osteoid osteomas believed to reach 15—20 mm in diameter at the most and osteoblastomas to have the potential to become considerably larger.

The current consensus is that osteoid osteoma and osteoblastoma are two distinct entities that differ regarding their topographic distribution, clinical and radiological presentation and, above all, potential for progression [6—9]. However, reports of borderline forms that are difficult to classify and descriptions of a few cases of osteoid osteoma believed to have transformed into osteoblastoma have given new impetus to the old single-tumour concept [10—12].

Here, we present a detailed analysis of two cases that seemed consistent with transformation of osteoid osteoma into osteoblastoma. Whether these two tumour types are of the same lineage is discussed.

Case report #1

An 11-year-old girl experienced gradual onset of pain at the lateral aspect of her left knee, with no precipitating factor. The pain was mechanical initially then became continuous (without nocturnal awakenings). Radiographs obtained 2 weeks after symptom onset showed a lucency measuring 12 mm along its long axis and located in the anterolateral cortex of the proximal metaphysis of the left tibia (Fig. 1). By computed tomography (CT), the lucency measured 9 mm and exhibited a sclerotic rim and a calcification suggestive of a nidus (Fig. 2).

Under the hypothesis of an osteoid osteoma, a radionuclide bone scan was performed. Marked uptake predominating in the early phases and persisting in the late phases was seen at the site of the lesion (Fig. 2).

These findings were consistent with osteoid osteoma. Given the location of the tumour, open excision surgery was scheduled. The pain became more severe and continuous, with a lancinating quality and loss of the response to paracetamol. Aspirin was introduced and alleviated the pain for several weeks. Aspirin therapy was stopped 2 weeks before the scheduled date of surgery, and the pain recurred gradually during this period.

The patient was admitted 5 months after symptom onset for surgical removal of the tumour. Intraoperative fluoroscopy to identify the lesion showed considerable expansion of the lucent defect, which measured 30 mm along its long axis. Consequently, a biopsy was performed instead of tumour excision. Findings during the biopsy were disappearance of the lateral cortex in contact with the lesion and thickening of the periosteum. The biopsy specimen was soft, fleshy, friable, and reddish in colour, with small foci of white granulations. It was sent to the laboratory for routine histological and bacteriological studies.

Radiographs and CT were performed immediately after the bone biopsy (Fig. 3). The bacteriological studies were negative. Histology showed an aggressive lesion, about which the advice of external experts was sought (Timone Marseille Teaching Hospital, France; and Mayo Clinic, Rochester, MN, USA). The final diagnosis was aggressive osteoblastoma rather than low-grade osteosarcoma (or osteoblastoma-like osteosarcoma) (Fig. 4 and 5). The surgical treatment consisted in painstaking curettage and filling of the defect with autologous bone. Histological examination of the residual tumour tissue removed during this procedure indicated osteoblastoma, with a less aggressive behaviour than in the biopsy specimen.

The pain resolved completely after surgery. At re-evaluation 6 months later, the patient was symptom-free despite having resumed normal sporting activities, and her radiographs showed bone remodelling at the surgical site.