

## Osteoid osteoma with a multicentric nidus

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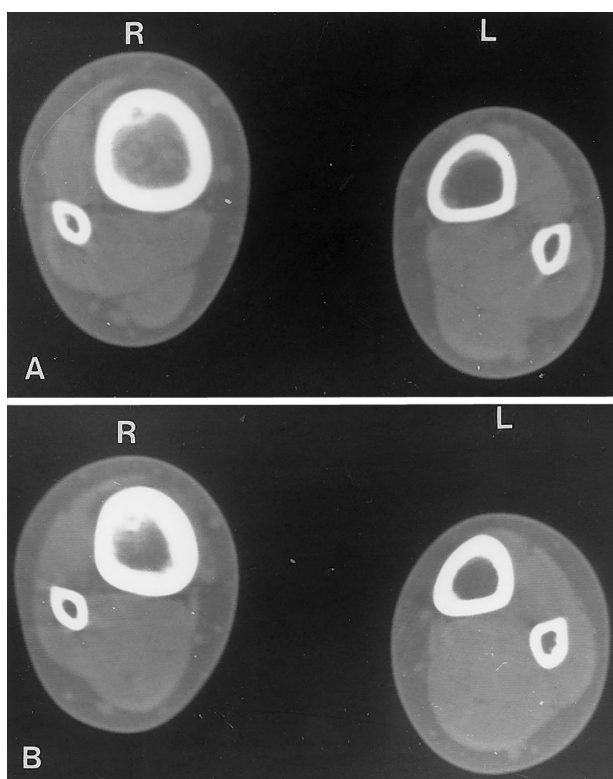
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**Summary.** A case is reported of a 16-year-old boy who presented with continuous pain in his right leg. Cortical thickening and diffuse medullary sclerosis was revealed on x-ray of the distal tibia. CT imaging showed a circumscribed annular pattern extending some 2.5 cm-s and indicating the multifocal nature of the lesion. The diagnosis of multifocal osteoid osteoma was confirmed after histological examination of the block of resected bone.

**Résumé.** On présente le cas d'un enfant âgé de 16 ans qui présente une douleur continue de la jambe droite. L'examen radiographique montre un épaississement de la jambe droite et une sclérose médullaire diffuse du tibia au tiers distal. L'image CT montre un épaississement annulaire circonscrit présent sur différentes sections espacées de 2,5 cm et nous suggèrent la nature multifocale. La résection en bloc et l'examen histologique confirment le diagnostic d'ostéome ostéoïde multifocal.

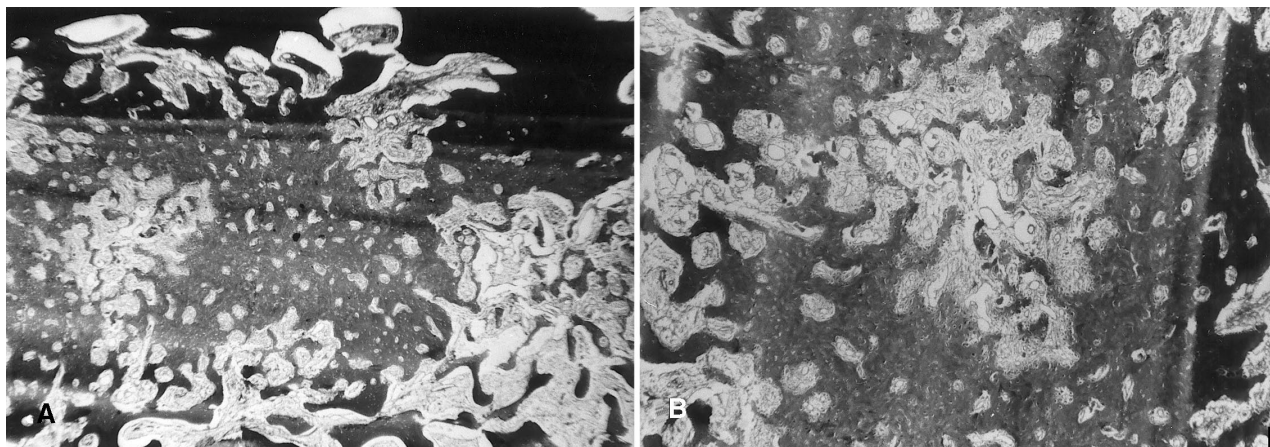
### Introduction

Multifocal osteoid osteoma is an uncommon type of osteoid osteoma. It was originally defined by Schajowicz in 1970 by the presence of “more than one circumscribed lesion, of a size similar to that of a central nidus of osteoid osteoma, all enclosed in a single block of sclerotic bone” [6]. Since then little more than a dozen such cases have been re-

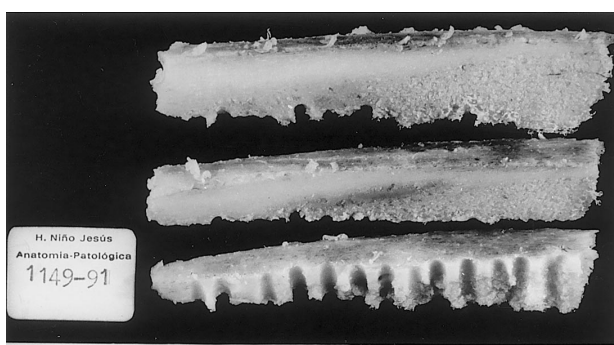


**Fig. 1 A, B.** CT sections 2.5 cm apart show the multicentric topography of the lesion and reveal a ring-like structure consistent with the “nidus” of an osteoid osteoma

ported, although it is possible that in others labelled as “osteoid osteoma”, the multifocal character was missed.



**Fig. 2.** **A** Juxtacortical multifocal osteoid osteoma with three nidi, surrounded by sclerotic bone (230 $\times$ ); **B** Nidus of highly vascularized osteoid tissue (230 $\times$ )



**Fig. 3.** Macroscopic fragment. A 28 mm long brown-coloured area can be identified. Extensive bone sclerosis and cortical thickening are observed

### Case report

A 16-year-old boy presented in the Outpatient Clinic complaining of continuous pain and swelling in the right leg for two months. He did not require analgesics because the pain was mild. On physical examination there was no reddening, heat or tenderness. His legs were otherwise normal.

Laboratory studies, including blood cell counts, erythrocyte sedimentation rate and routine blood chemistry were normal.

Plain radiographs showed a diffuse increase in bone density with cortical thickening of the distal right tibia. A CT scan showed thickening of the anterior cortex and a ring-like intracortical structure extending 2.5 cm-s along the tibial diaphysis (Figs. 1a and 1b). A wide block resection (7 cms  $\times$  1.5 cms) of the lesion was performed (Fig. 3). In this specimen there was a macroscopic lesion, 2.8 cms long and brown in colour. It contained multiple osteoblastic foci with osteoid trabeculation within highly vascularized connective

tissue, the whole surrounded by reactive sclerotic bone tissue. This was considered to be a multifocal osteoid osteoma.

### Discussion

In the literature we have found neither precise nor clear clinical differences between the presentation of single and multiple osteoid osteoma [3]. Pain is generally the major clinical symptom in both types [3] and frequently it is worse at night. Small doses of salicylates may give relief [2].

The clinical presentation will depend on the localization of the lesion. If subperiosteal the nidus may extend into the soft tissues raising the periosteum [4, 6]; a juxta-articular tumour may provoke synovitis [3, 5] or if near the physis may result in accelerated growth.

We have not found any differences in the physical findings between the two types of osteoid osteoma.

The histological features of our case fit Schajowicz's criteria of multifocal osteoid osteoma. The pathogenetic significance of this lesion and the relationship between conventional osteoid osteoma and osteoblastoma are unknown. Some authors suggest that multifocal osteoid osteoma is an intermediate entity between these two conditions.

Multifocal osteoid osteoma has been observed in locations which are unusual for conventional osteoid osteomas, such as the ethmoid bone, the pubis [6], and the fingers [3]. The tibia is a common site for both the conventional and the multifocal forms, but the radiological appearance is different. Sometimes it resembles a cortical fibroma [5, 7]. Plain photographs may show a

characteristic nidus and the multifocal nature [2]. In our case, extensive sclerosis obscured all other details, and only the CT scan revealed its multifocal form. The excised bone fragment contained an elongated dark area, greater than that normally found in osteoid osteoma, and microscopical examination confirmed the presence of several distinct osteoid lesions (Fig. 2).

The final diagnosis of multifocal osteoid osteoma is made by the pathologist who requires a well-defined non-fragmented sample. Block resection is therefore preferable to curettage when technically feasible.

Wide resection is also advantageous because the multifocal condition of a lesion could theoretically be related to its possible recurrence. Unfortunately, recurrence is not uncommon in osteoid osteoma [5], and is traditionally attributed to incomplete removal. It has been suggested that in some cases of “recurrence” of osteoid osteoma their original multifocal nature had been missed [1].

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