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CLINICAL COMMENTARY

Benign osteoblastoma in an unusual mastoid location

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KEY WORDS
Osteoblastoma;
Bone tumors;
Mastoid;
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Histology

Summary
Introduction: Benign osteoblastoma (OB) is an unusual primary bone tumor. The preferred locations are the posterior arch of vertebrae and long bones. We report herein an extremely rare location of an OB in the mastoid process of the temporal bone.
Case report: A 22-year-old woman presented with painful left retro-auricular swelling. Computed tomography features were suggestive of an aggressive osteolytic lesion of the left mastoid. The pathologic examination of bone curettage material revealed a benign OB. A complete resection of the tumor was performed later, with no evidence of recurrence at 1 year.
Discussion/Conclusion: To our knowledge, this is the 14th reported case of OB confined to the mastoid process of temporal bone. Its histological diagnosis can be difficult and osteosarcoma is its principal differential diagnosis. Although generally regarded as benign, OB has potential for recurrence and local invasion. As such, complete resection, whenever possible, is preferred over conventional curettage.
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Introduction

Osteoblastoma (OB) is a rare, benign primary bone tumor, most often occurring in the posterior arch of vertebrae and the long bones of the lower limbs [1—3]. Its location in the temporal bone is rare. We report a new case of OB located in the mastoid process of the temporal bone.

Observation

A 22-year-old female patient with no significant past medical history, presented in January 2008 for left-sided otalgia, which had been evolving over 2.5 years following temporal trauma. Pain was isolated with no associated otorrhea or dizziness, hypoacousia, or neurological signs. This pain had been progressively intensifying until 2 months before consultation, at which time left retroauricular tumefaction appeared.

Examination showed sensitive retro-auricular swelling facing the superior part of the mastoid. No associated neurological deficit or reduction in auditory acuity was observed.

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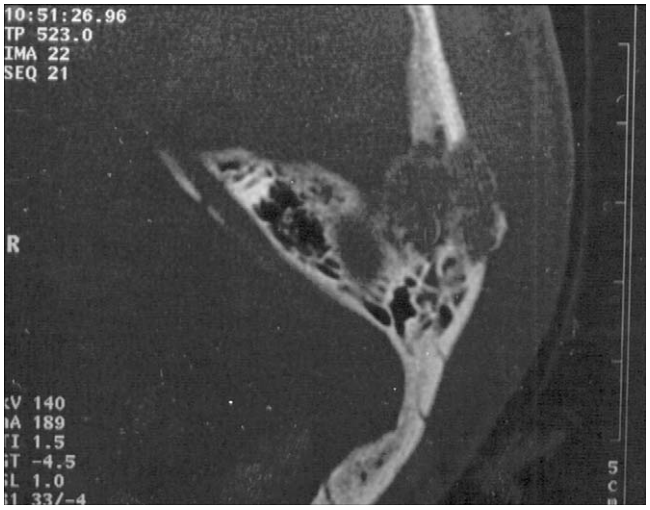

Figure 1 Axial CT scan of temporal bone showing a 4-cm lytic lesion of the left mastoid extending to the temporal squama.

Computed tomography (CT) of the petrous part of the temporal bone showed a left-sided 4-cm mastoid lytic process, extending upward toward the squamous temporalis, involving the outer and inner tables of the bone (Fig. 1). Given these clinical and radiological aspects, the diagnosis of a tumoral process was judged to possibly be a Langerhans histiocytosis.

Bone curettage of diagnostic intent was performed. Microscopically, the product of this curettage showed a benign bone-forming tumor composed of a network of osseous trabeculae at variable stages of maturity within a highly vascular stroma (Fig. 2). These trabeculae were lined by one or several rows of active osteoblasts, with scattered associated osteoclasts (Fig. 3). These findings were consistent with benign OB.

The patient was lost to follow-up and was seen again only in January 2009, with a lesion that appeared stable from a clinical and radiological points of view. The patient underwent surgery in June 2009, with ablation of the tumor, which had extended to come in contact with the dura mater, with reaming of the healthy bone margins. Histological examination of the gross specimen confirmed the diagnosis of OB. Postoperative recovery was uneventful and the patient was well without any sign of recurrence 1 year after surgery.

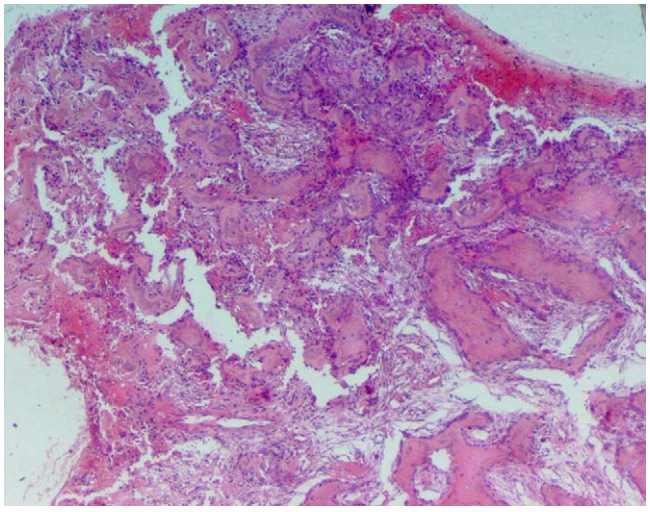

Figure 2 (H&E × 100): trabeculae at varying stages of maturity.

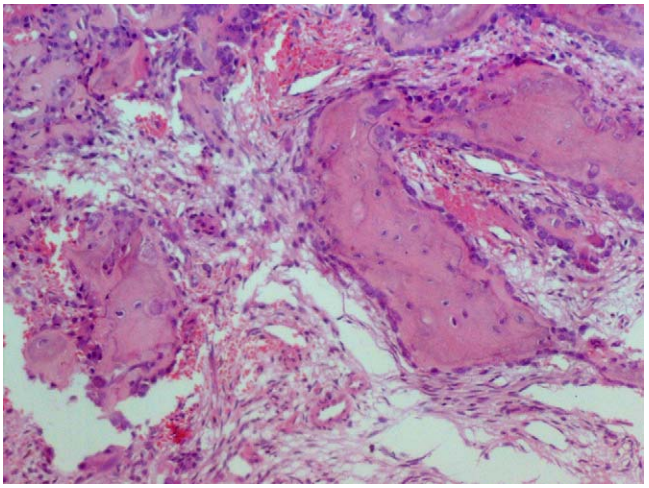

Figure 3 (H&E × 200): trabeculae surrounded by two or more rows of active osteoblasts.

Discussion

OB is a rare bone-forming benign tumor, accounting for 1% of all bone tumors [4]. In the head and neck, it often involves the cervical vertebrae, the maxillary bones, or the frontal bone. Location on the temporal bone is rare and on the mastoid process even rarer [2,4]. To our knowledge, the case reported herein is the 14th OB of the mastoid described in the literature [3].

In its usual locations, OB is more frequent in male subjects aged between 10 and 30 years (90% of cases). However, a clear female predominance has been noted in head and neck locations, particularly with the temporal bone [3—5]. The mean age at onset is 20 years (range, 7 months to 61 years) [3].

Symptoms include tumefaction associated with continuous bone pain that are not relieved by minor antalgics, more rarely by tinnitus, reduced auditory acuity, or facial paralysis following involvement of the adjacent cranial nerves [3,6,7].

The radiological appearance of the tumor is that of an intramedullary, round or oval, well-demarcated lytic lesion, with varying degrees of calcification [6]. MRI shows an iso- or hypointense signal on T1-weighted sequences and a hyperintense signal on T2-weighted sequences [2,3,5]. In some forms of aggressive OB, the radiological appearance is very misleading and can mimic osteosarcoma, especially in flat bones [8]. In cases of intracranial development, the radiological differential diagnosis with meningioma can be difficult [5].

Macroscopically, OB is a red tumor generally measuring more than 2 cm in diameter, with cystic areas clearly delimited from the adjacent bone [7]. Histologically, this tumor comprises randomly distributed trabeculae

of variable maturity, surrounded by two or several rows of osteoblasts. These trabeculae are separated by richly vascularized stroma with multinuclear giant cells of the osteoclastic type [4]. Mitoses are very rarely observed within an OB. The presence of large osteoblasts containing nuclei with prominent nucleoli defines epithelioid OB. The biological significance of this variant of OB is still disputed. Some authors associated it with a more aggressive clinical course. Other authors find no such correlation; today there exists no predictive histopathological criterion of the clinical behavior of OB [1,4,7]. The lesion can include areas of secondary aneurysmal cysts [4]. The presence of cartilage within an OB, except in case of associated fracture, is exceptional and should first and foremost rule out the possibility of osteosarcoma, which constitutes the main histological differential diagnosis of OB. Findings against the diagnosis of osteosarcoma include the absence of abnormal mitosis, cytonuclear atypia, and permeation of the surrounding intertrabecular spaces. The distinction may sometimes be extremely difficult and requires correlation with the radiological features [1,5]. More rarely, OB may mimic ossifying fibroma or osseous dysplasia.

Even though OB is a benign tumor, most authors recommend ''conservative'' complete resection and prefer it over curettage. This resection is sometimes difficult given the narrow anatomical spaces between the tumor and the adjacent neural structures. Preoperative arterial embolization is advocated by some authors so as to reduce bleeding during surgery [3,4,6]. Radiotherapy is increasingly set aside because of the risk of transformation into osteosarcoma [7]. The outcome after treatment is generally favorable with the exception of rare locally aggressive forms. Local recurrence was noted in five of 31 (16%) patients with follow-up time to recurrence varying from 7 months to 11 years after resection [5].

Conclusion

OB of the temporal bone, and in particular, of the mastoid, is very rare, making its preoperative diagnosis very difficult. It most often affects young females and presents as a slow-growing painful lump. Conservative treatment is sufficient in most cases with a good prognosis.

Conflict of interest

None.

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