

CASE REPORT

Aneurysmal Bone Cyst: An Unusual Benign Lesion of the Temporal Bone

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Aneurysmal bone cyst are osteolytic lesions of the long bones that rarely occur in the temporal bone. Successful therapy focuses on complete tumor removal to avoid recurrences. The unusual case of a 15 year old patient with a rapidly growing tumor involving the temporal bone is presented. The typical features of this lesion on CT scans, the surgical removal and follow-up is discussed.

Submitted : 05 November 2012

Revised : 01.05.2013

Accepted : 06.05.2013

Introduction

Aneurysmal bone cysts (ABC) were first described in 1942 by Jaffe et al. and constitute a benign, rapidly growing osteolytic lesion consisting of two parts: an intraosseous, destructive tumor and an extraosseous, aneurysm-like cyst.^[1] This non-neoplastic lesion of unknown origin presents mostly in metaphyses of long bones of adolescent patients. The presentation in the skull either with extra- or intracranial extension is rare with an incidence of 3 - 6%. A high level of recurrence up to 40% has been described in non-skull ABC.^[2] Neurological symptoms such as facial nerve paralysis have also been reported in temporal bone lesions as result of intracranial tumor compression^[3].

This article presents one of the few known cases of an extensive ABC of the squama and zygoma of the temporal bone without intracranial extension; the clinical properties, radiologic features and surgical approach are discussed together with the relevant literature.

This study was approved by the Ethics committee of the Kantonsspital Luzern on August 31st, 2012.

Case Report

A 15 year-old girl was referred to our clinic due to the rapid growth of a soft tumor behind the left ear within the last three months; this lesion was first observed two years earlier, but neglected due to its painless nature and initial small size. The patient had suffered from few episodes of uncomplicated recurrent acute otitis media during early childhood and was otherwise healthy.

The clinical examination showed a 5cm round, soft, "air-filled", slightly tender, well circumscribed temporo-parietal tumor displacing the pinna antero-caudally. A Valsava maneuver was avoided after being informed, that variations in size were noted after sneezing.

A high resolution computed tomography (HR-CT) of the temporal bone showed a well aerated trabecular

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bony expansion arising from the left squama, exteriorizing subcutaneously and reaching the zygomatic bone (Figure 1-A). A normal anatomy of the middle and inner ear was verified and the hearing tests were normal.

Surgical Procedure

A ventilation tube was placed in order to decrease pressure while sneezing and for the preparation of the removal. After exposing the mastoid plane, a thin convoluted bony lamella between the temporal line and the antrum was identified. This lesion appeared to be firmly adherent to the periosteal fibers (Figure 2). This trabecular bone formation extending between the zygomatic arch, the squama temporalis and the squama occipitalis was drilled away until reaching the tabula interna of the temporal bone, skeletonizing the dura of the tegmen tympani and partially removing bone from the zygomatic arch. The antrum was left unexposed and few superficial cells were filled with bone-wax and cellulose strips (Tabotamp[®]). Periosteal tissue and the temporalis muscle were reattached and the skin closed. An easy-flow-drainage was removed after 2 days. The postoperative period remained uneventful and the ventilation tube spontaneously extruded few months later.

Histopathological examinations showed the typical areas of extravasated blood in bony cavernous spaces and the presence of giant cells without signs of malignancy. These findings were consistent with the diagnosis of an ABC.

Follow-up

The patient was regularly controlled postoperative and no complications were noticed. The clinical examination and HR-CT 12 months after surgery were unremarkable showing complete removal of the lesion without signs of recurrence (Figure 1-B). The patient had no further complaints.

Discussion

We present one of the few known cases of aneurysmal bone cyst in the temporal bone. According to previous publications and similarly to our case, 80% of the patients, predominantly females, present with externally visible lumps, often tender on palpation within the first 2 decades of life [4]. Neurological symptoms have been described depending on the location and extend of the lesion.

HR-CT was sufficient to provide a preliminary diagnosis showing a well aerated lesion in the squama

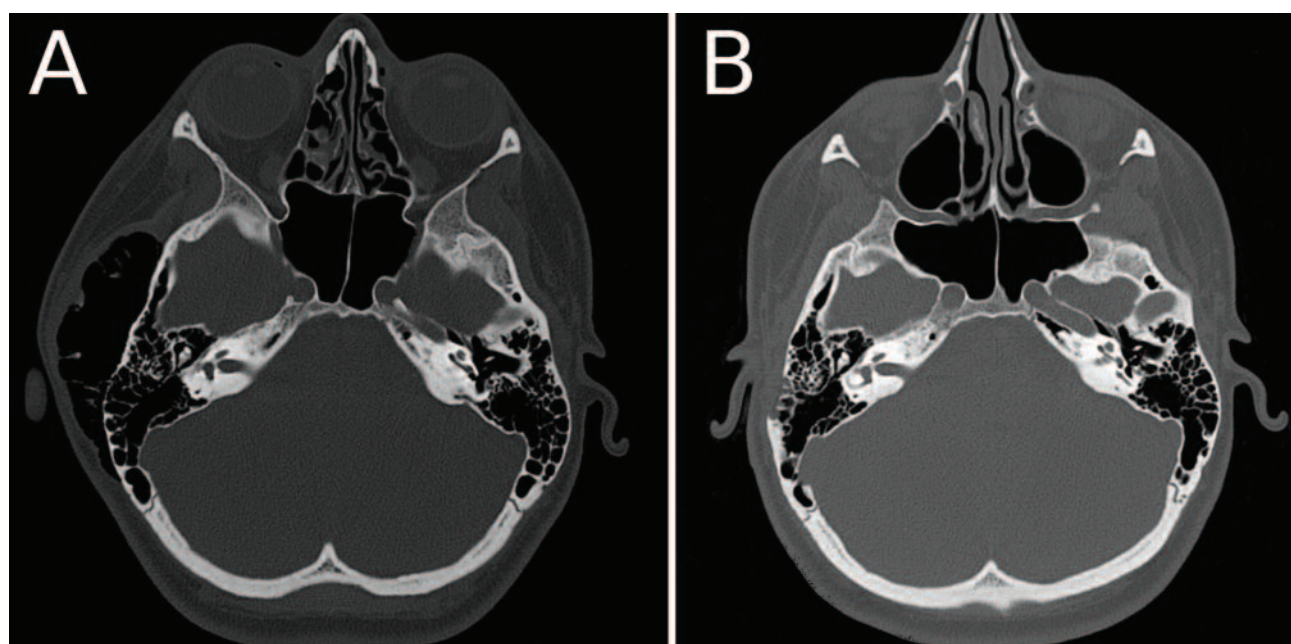


Figure 1. HR-CT of the temporal bone showing the extensive trabecular cystic bone formation along the squama and zygoma before (A) and after complete cyst removal 12 months later (B).

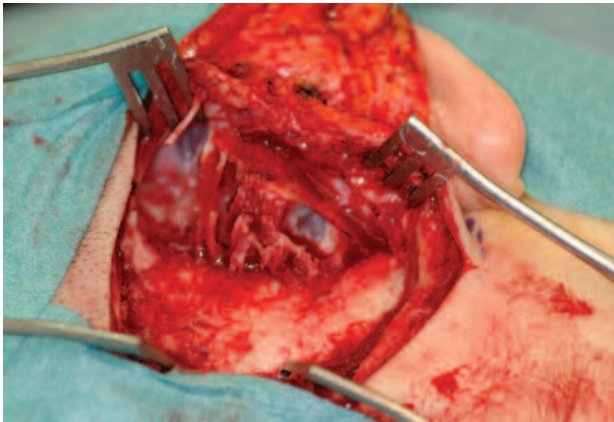


Figure 2. Intraoperative view of the trabecular cystic bone formation in the subperiosteal-subcutaneous tissue after exposing the mastoid plane.

of the temporal bone extending superficially toward the zygomatic process without signs of hypervascularity or cystic lesions. ABC may also present as heterogeneous lesions with multiple fluid filled cavities, internal septations and a high hypervascularity. These features are better analyzed on MRI to exclude other pathology^[5,6].

Although most of the ABC occur de novo, in other instances it may correlate to previous trauma or coexisting bony lesions such as preexisting chondroblastoma, osteoblastoma, giant cell tumor, fibrous dysplasia or a malignant osteosarcoma.^[7] The risk of recurrence and the association with malignant tumors mandate a complete removal of the lesion. Due to the rarity of this benign tumour, there is no consensus regarding the recurrence rate for temporal ABC but a high risk has been reported from non-skull lesions. The successful use of local radiotherapy to a dose of 31.5Gy has been reported in a patient with a recurrent, large ABG of the temporal bone^[8]. In our case the lesion could be completely removed and after a year of follow-up, no

complications or signs of recurrence were observed in the HR-CT.

We conclude, that early clinical suspicion and verification by CT scanning (and MRI in case of heterogeneous presentation) should enable the surgeon to completely excise this superficial temporal bone lesion and avoid recurrences.

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