Acta Scientific Otolaryngology (ISSN: 2582-5550)

Volume 3 Issue 9 September 2021

Case Report

Aneurysmal Bone Cyst of the Maxilla: Report a Case

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Received: July 23, 2021

Published: August 09, 2021

Abstract

An aneurysmal bone cyst (ABC) is a rare benign multicystic mass, which its frequency in the head and neck is very low. The majority of ABCs of head and neck are located in the mandible and there are few cases with ABCs in the maxilla bone. It is commonly found in patients younger than 20 years old. ABC should be considered in a differential diagnosis of benign swelling in the sinonasal area. Here we present the case of a young woman with a massive aneurysmal bone cyst of the anterior skull base as an educational example and a reminder to consider ABC in the differential diagnosis of benign swelling in the maxillofacial area.

Keywords: *Aneurysmal Bone Cyst; Maxillofacial, Spiral Computed Tomography; Sphenoid Sinuses*

Introduction

An aneurysmal bone cyst (ABC) is a rare benign multicystic mass that constitutes 1 - 2% of whole primary bone marrow masses [1]. The frequency of ABC in the head and neck is very low, accordingly, ABC of skull and facial skeleton is reported in 3 to 6% of all these masses [2]. In addition, of the majority of ABCs of head and neck (66%) are located in the mandible and there is few cases with maxillary ABCs [3]. No gender predilection has been reported for ABC [1]; however, epidemiological variation in different parts of the world has been reported in the literature [4]. The majority of the case with ABC is reported in the Brazilian studies [5].

ABC is commonly found in patients younger than 20 years old. They occur usually as primary bone tumors or secondary to malignancy trauma or vascular dysfunction. These tumors usually involve metaphyses of tibia and femur and vertebrae and flat bones [6].

Here we present the case of a young woman with massive aneurysmal bone cyst of anterior skull base as an educational example and a reminder to consider ABC in the differential diagnosis of benign swelling in the maxillofacial area.

Case Report

A 13 year old female with a severe swelling in maxillofacial area and left progressive proptosis and a protruding mass from left nose was referred to our center. The patient complained of diplopia with visual acuity was normal. According to the patient the swelling was growing very fast with accompanying epistaxis and nasal obstruction from 2 months ago the swelling was nontender otherwise. The general health of the patient was normal with no cervical lymphadenopathy. Left ocular movement was decreased and medial movement of globe was limited. chest radiography and blood analysis were normal. Figure 1 presents the spiral computed tomography (CT) scan of the of the *paranasal* sinuses.

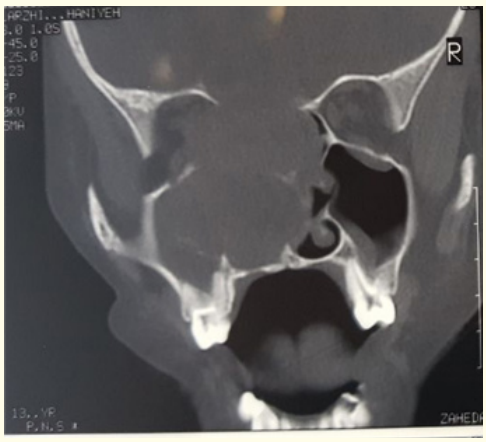

Figure 1: Spiral computed tomography (CT) scan of the of the paranasal sinuses.

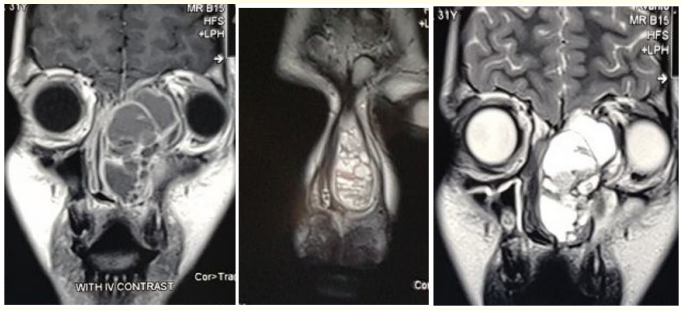

Figure 2: Coronal T2-weighted and contrast enhanced T1-weighted MR image from skull base show an expansile multiseptated cystic mass with fluid-fluid level in T2 and septal enhancement in post contrast image.

The magnetic resonance imaging has revealed expansile multiseptated cystic mass with fluid-fluid levels, as it presents in the figure 2, which showed extension in the medial wall of the left orbit, nasal cavity and left maxillary sinus with mass effect without invasion and destruction also seen in CT scan images. In addition, here was no invasion to the brain.

The patient operated with endoscopic assisted Denker approach and the whole mass was resected macroscopically. During the surgery, the pathology was mainly in the right nasal cavity, grossly involving the cribriform plate and the medial orbital roof. The pathology was totally extradural, and the bone had undergone substantial transformation with shift of the right globe laterally. An extremely thin osseous layer was meticulously separated from the dura. The pathology encircled numerous cystic parts having gloomy fluid due to longstanding blood products. Connection of the dura to the crista galli was obstructed and separated. The resection stretched down to the ethmoid cells, and the maxillary and sphenoid sinuses. The diagnosis of ABC was confirmed by a histopathological examination. The pathology revealed blood filled cystic lesions containing osteoclast, as it shown the figure 3.

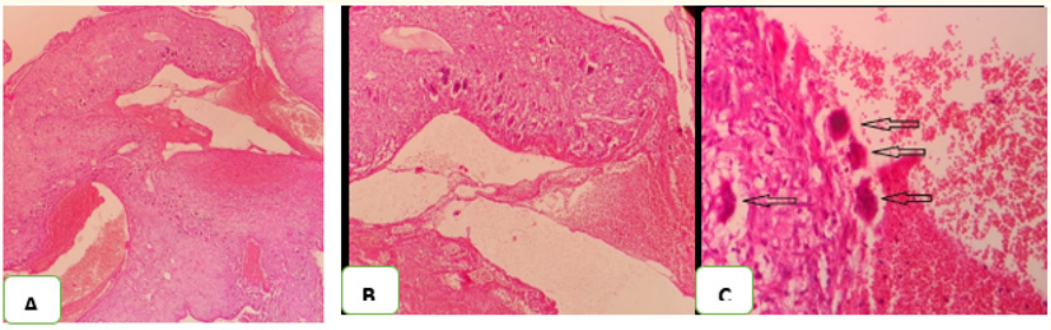

Figure 3: Blood filled cystic lesions (Haematoxylin and eosin ×25) was observed in histopathology (A), blood filled cysts with fibrous septa, no epithelial lining (Haematoxylin and eosin, × 100), photomicrograph of the cyst wall shows C fibrous cyst wall containing osteoclast like giant cells (arrows) (Haematoxylin and eosin, ×400).

The patient had an uneventful recovery and was discharged on the postoperative day 10. Her proptosis had resolved and vision was good at the 3-month follow-up. Neurologic examination was normal, and the patient reported a good general condition.

Discussion

This article presents a rare case of ABC in the maxillary bone. ABC was recommended based on radiological and pathological findings that were compatible with surgical findings.

The origin and precise nature of ABC are unknown. The mass may be primary or secondary to another skeletal lesion [7]. A review of Mankin., *et al.* has been described in 150 ABC patients. The mean age was 18 years old. ABC is frequently related to intermittent headache and nasal block. The severity of the symptoms is determined by the position and size of the pathology [8]. There have been former reports of ABC in different parts of head and neck. But although it is felt that these lesions are benign, our case shows that they compress rather than invade neighboring structures. It is felt that the symptoms of diplopia and headache are due to the expansile nature of the tumor.

ABCs may be primary or secondary, with primary lesions appearing in the isolation and secondary lesions in the environment of the previous or consequently damaged bone marrow. The most common lesion after this is a giant cell tumor. Other lesions include chondroblastoma, osteoblastoma, osteosarcoma, nonossifying fibroma, and fibrous dysplasia [9]. It seems ABCs are always a secondary phenomenon in a pre-existing lesion, which already affects the involution changes, triggers an intravenous arterial abnormality and, through hemodynamic forces, creates a secondary bone reaction that we have identified as an aneurysmal bone cyst. A previous lesion may be detected in about one-third of the ABCs histopathologically and may be completely obliterated by ABC in cases where no lesion has been detected [10].

Due to rarity and overlapping radiological features of ABC, it is difficult including this pseudocyst as the initial clinical hypothesis [5,11]. Histopathological diagnosis of ABCs is difficult because of the association of ABCs with other bone tumors and their similarity with some microscopic features of simple bone cyst, central giant cell lesion, and benign fibro-osseous lesions [12,13].

ABC may be misdiagnosed by other bone pathologies because of radiographic appearance of "ground glass" classic [4]. Although it is possible that ABC is considered as a tumor to angiofibroma; the clinical behavior of the tumor facilitates its diagnosis. The main characteristic of ABC is hemorrhagic fluid content and septated appearance [2]. Commonly, the benign multicystic mass is expanded rapidly and locally destructive with a clear characteristic in Computerized findings (CT) and Magnetic resonance imaging (MRI) [3].

The main findings in CT include erosion, thinning of cortex, bony ridges, and the expansion of Diploic spaces, bone widening, ground-glass opacity, and increased contrast enhancement. There are fluid levels in CT in 30% to 35% of cases [6].

Magnetic resonance imaging (MRI) is more than CT scan to show the level of the fluid, especially in T1 images, as well as the presence of a solid component that indicates which ABC is the secondary one. In this study, we found well-defined heterogeneous signal intensity lesion on T1, as it is common in the other cases with ABC. Hypointense capsule and a homogenous enhance in signals intensity on T2 is the other findings of ABC. Cysts have variable signal, with margins have low T1 and T2 signals. Focal areas of high T1 and T2 signals are seen presumably representing areas of blood of variable age. It is important to remember that the presence of fluid levels, although the characteristics of ABC, are by no means unique to them and are seen in other lesions, both benign and malignant (e.g. giant cell tumors (GCT), chondroblastoma, simple bone cysts and telangiectatic osteosarcomas) [14,15]. Due to the aforementioned data, clinical and imaging aspects and representative biopsy specimens should be the correlated to diagnosis of ABC.

The treatment choice for ABC of the skull is complete resection that may be difficult when the lesion is large or involves the base of the skull. In this study surgical excision was performed considering typical CT and MRI findings. Because of the rarity of ABC in the maxilla, all treatment options including sclerotherapy, embolization, and radiotherapy should be considered. Preoperative intravascular embolization if the tumor is highly vascular is useful. Embolization is rarely used as a stand-alone treatment for lesions that are not surgically accessible. Radiotherapy has been used in some cases, but because of the high recurrence (higher than 30%) and risk of sarcoma degeneration, this treatment is reserved for irreversible or recurrent lesions and is rarely used. Total gross resection is generally curative, whereas topical or curettage resection has a recurrence rate of up to 50% [16]. The recurrence rate is reported in 10 to 30% of cases with total resection [8].

Conclusion

Although incidence in the anterior skull base is rare, ABC should be considered in a differential diagnosis of benign swelling in the sinonasal area.

Conflict of Interest

There is no conflict of interest.

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