Primary aneurysmal bone cyst of maxillary sinus-report of a rare case

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Citation

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Abstract

Aneurysmal bone cyst (ABC) is a non-neoplastic expansile bone lesion that is common in the long bones. Only 2% of these occur in the head and neck region. Mandible is usual site of involvement in facial skeleton Their occurrence in the maxilla is rare. Our case report describes a primary aneurysmal bone cyst (ABC) of the maxillary sinus in a 18-year-old female. The patient presented with progressive, rapidly growing swelling of the left cheek. Imaging studies showed a heterogeneous contrast enhancing mass expanding the left maxillary sinus. The lesion was resected endoscopically and histological examination revealed aneurysmal bone cyst.

INTRODUCTION

Aneurysmal bone cyst (ABC) is described as a localized and quickly expansile benign tumor, which can reach a considerable size. ABC is very rare among all the cystic lesions that can be found at the mandible or the maxilla[1,2]. We report the CT and morphological findings of ABC of the maxillary sinus in a 18 year old female. It adds one more aspect in diversity of clinical and biological behavior of ABC in maxillofacial region.

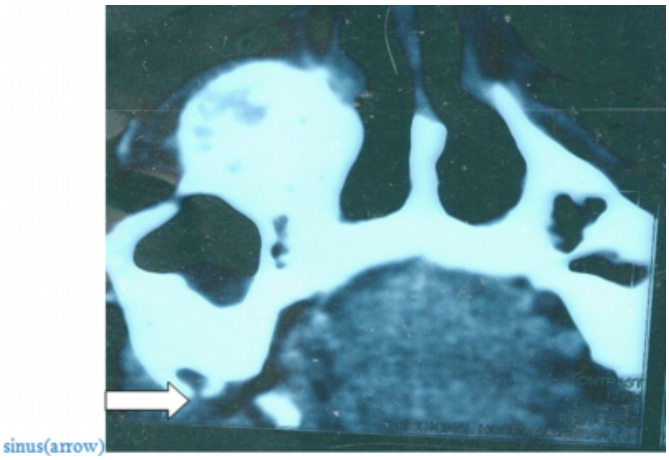
CASE REPORT

A 18 year old female patient presented with a progressively increasing swelling over the left cheek, which developed over the previous 8 months. There was mild pain on pressing the swelling and on opening of mouth. Swelling was progressively increasing in size. There was no other complaint

On examination, there was about 3 × 3 cm bony swelling in the left cheek. It was immobile, slightly tender with egg shell crackling on pressure. There was no murmur or bruit on auscultation. On aspiration, brownish serous fluid came out. It contained mainly red blood cells. CTscan of head and face (Figure 1) revealed rounded expansile lesion of bone with cortical thinning ,originating from left maxilla(arrow).The lesion showed multiple internal septations and cysts with fluid-fluid levels of varying intensity(Fig1)

Figure 1

Figure1: CTscan of head and face revealed rounded expansile lesion in the left maxillary



The tumor was excised endoscopically and histopathological examination (Figure 2) showed numerous variably sized blood filled cystic spaces(arrow) separated by fibrous septae containing spindle shaped fibroblasts and scattered multinucleated giant cells(Figure3).

Figure 2

Figure2: Photomicrograph showing cystic vascular spaces (arrow) separated by septa containing giant cells and fibroblasts. (H&E 250×)

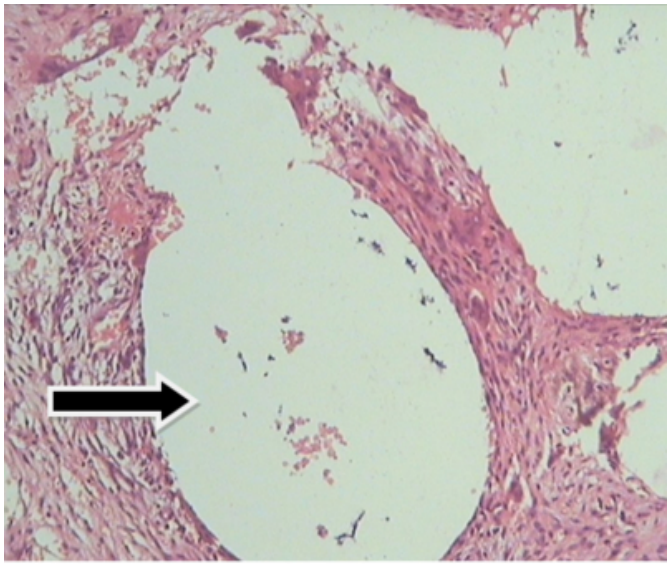
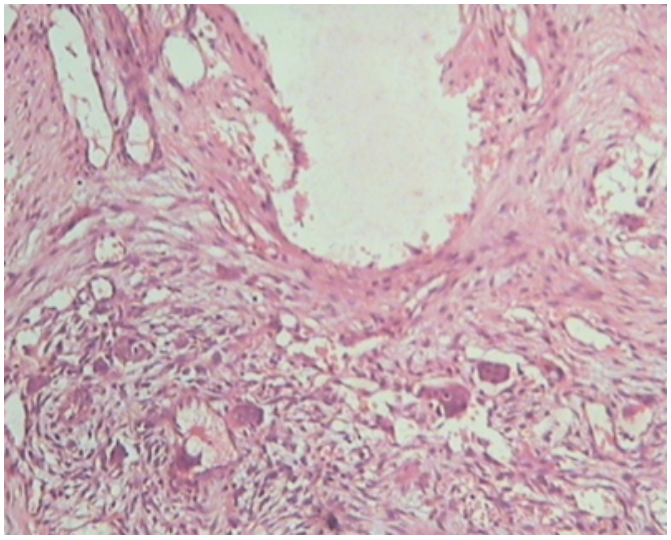


Figure 3

Figure3: Higher magnification showing osteoclast type of giant cells and proliferating fibroblasts in the wall (H&E,400x)



DISCUSSION

Aneurysmal bone cyst (ABC) is a expansile benign bone lesion that is common in the long bones; only 2% occurs in the head and neck region[1].It rarely presents in the maxillary region[2,3]

Aneurysmal bone cyst can reach a considerable size. It is neither an aneurysm nor a true cyst, but rather a non-neoplastic, osseous lesion characterized by the presence of numerous blood-filled, usually nonendothelialized cavities. According to WHO, ABC is an “expanding osteolytic lesion consisting of blood-filled spaces of variable size separated by connective tissue septa containing trabeculae of osteoid tissue and osteoclast giant cells.[4]

Aneurysmal bone cyst is most common in those regions of the skeleton where there is both a relatively high venous pressure and high marrow content [5]. This explains rarity of ABC in the skull bones in which there is low venous pressure. It is attributed to a circulatory disturbance leading to locally increased venous pressure but with an unclear etiology.[6] It takes place mainly in the long bones and in patients less than 20 years old, with no sex predilection. It can attain great dimensions and may cause symptoms owing to its site and size and rapidity of growth i.e. swelling, deformity, pain, neurologic symptoms, and pathologic fractures.While CT (and MR) demonstrate characteristic fluid-fluid levels, the diagnosis remains histological[7].

It can occur as a primary lesion or secondarily in a preexisting lesion. Giant cell tumor is the most common lesion associated with secondary ABC accounting for 39% of these lesions and similarly in 14% cases of giant cell tumor, ABC components are seen. The other associated lesions are unicameral cyst, nonossifying fibroma, osteoblastoma, hemangioma, histiosarcoma, hemangioendothelioma, fractures and trauma. Thus when treating ABC, it is important to determine whether any pre-existing lesion has preceded or not; in particular, if the lesion site is in the head and neck region[1].

There are various treatment options suggested in the literature ranging from per-cutaneous sclerotherapy, diagnostic and therapeutic embolization, curettage, block resection and reconstruction, radiotherapy and systemic calcitonin therapy. Self healing cases have also been reported on long term follow up [8].

ABC may behave aggressively and invade the orbit; so resection is necessary. Current recommended treatment is curettage with enucleation first if technically possible. Minimally invasive techniques such as endoscopic sinus[5] surgery can be performed successfully in select cases. Long follow up is important because recurrence may occur, in which case further resection is warranted.[9]

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