Case Report

Unusual case presentation of an aneurysmal bone cyst of maxilla in geriatric patient: A diagnostic challenge

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Abstract

An aneurysmal bone cyst (ABC) is a non-neoplastic, rapidly expanding pathologic bone lesion that mostly affects the mandible and is most commonly found in the first to third decade of life. The most of the ABC cases are locally aggressive osteolytic lesion with a high recurrence rate. As a result, we present a swelling with pus discharge at the maxillary alveolus in a 68-year-old female who was diagnosed with ABC after a computed tomography scan and histological analysis. The conservative surgical excision was performed without considerable bleeding, and no recurrence was observed even after a five-year follow-up period.

Keywords: Aneurysmal, bone cyst, CT, geriatric, histopathology, maxilla, surgical excision

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INTRODUCTION

An aneurysmal bone cyst (ABC) as defined by the World Health Organization is an expansive osteolytic lesion consisting of various size blood-filled spaces of channels which is separated by connective tissue septa that can contain multinucleated giant cells and osteoid tissue.[1] Van Arsdale was the first to recognise ABC in 1893, while Jaffe and Lichtenstein proposed the term "Aneurysmal bone cyst" in 1942 and explained that ABC is a pseudocyst that is not lined by epithelium. ABC is a relatively rare lesion of the head and neck region. Available literature shows that it most commonly occurs in long bones (50%), spine (12-30%), and jaws (2-12%). The lesion appears more common in the mandible than the maxilla, with a ratio ranging from 2:1 to 11:9 with onset in the third to fourth decade of life.[2] Here, we are presenting a rare case of ABC in a 69-year-old female patient at a maxillary alveolus.

CASE PRESENTATION

A 68-year-old woman presented with a two-month history of front upper gum enlargement, dull pain, and pus discharge. A history of facial trauma was given by the patient. The swelling extends from 23 to 13 cm intraorally, with 3 cm x 2 cm in dimension at the palatal and buccal cortex. On palpation, the soft tissue mass was reddish in colour, firm in consistency, and mild tenderness. [Figure 1] Contrast-enhanced computed tomography of maxilla showed a well-defined multiseptated expansile cystic lesion of upper anterior alveolus with thin out and resorption of remaining maxillary bony cortex [Figure 2]. The incision was carried out under local anaesthesia and sent for

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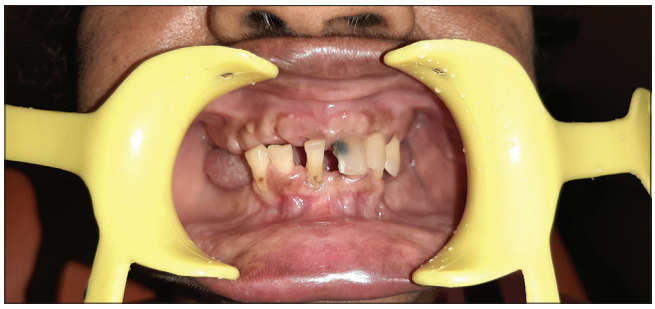

Figure 1: Preoperative photograph of the 68-yr-old female showing swelling in the anterior alveolar region of the maxilla

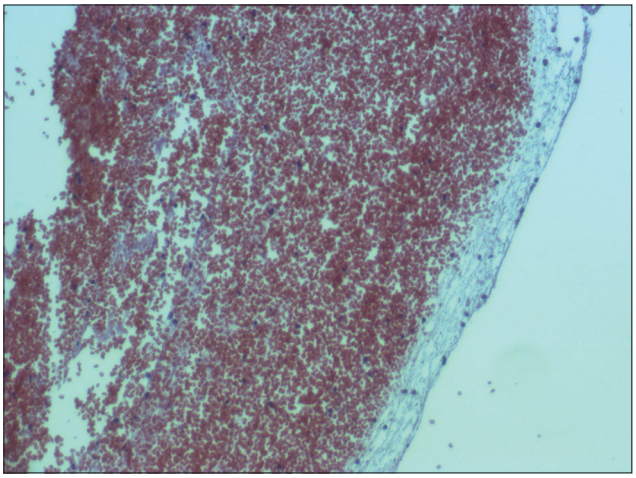

Figure 3: Histopathologic examination showing sinusoidal spaces filled with blood (H&E stain, ×400 magnification)

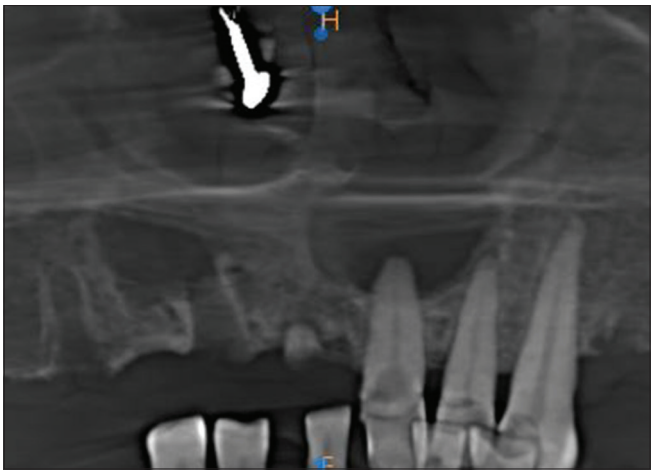

Figure 2: Computed tomographic scan of maxilla showing a multicystic expansile cystic mass in the maxilla

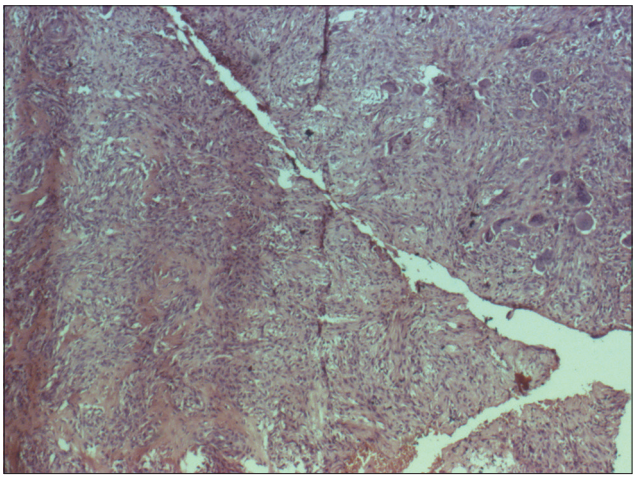

Figure 4: Histopathologic examination reveals multinucleated giant cells osteoid elements in fibrous tissue (H&E stain, ×400 magnification)

histopathological examination. On H and E examination, it revealed cavernous vascular spaces filled with blood and surrounded by fibrocollagenous tissue containing spindle-shaped cells, multinucleate giant cells, inflammatory cells, and reactive bone formation. The above clinical, imaging, and histopathological features were consistent with ABC. [Figure 3] Conservative surgical resection was carried out at the involved maxillary gingo-alveolar portion [Figure 4]. There was no excessive bleeding noted during surgery. It healed uneventfully and the patient is under regular follow-up for five years without clinical evidence of recurrence.

Under local anaesthetic, the incision was made and the tissue was sent for histological evaluation. On H&E-stained tissue section reveals cavernous vascular spaces filled with blood surrounded by fibro collagenous tissue including spindle-shaped cells, multinucleate giant cells, inflammatory cells, and reactive bone formation. The clinical, imaging, and histological characteristics identified above were all consistent with ABC. [Figures 3 and 4] The affected maxillary gingo-alveolar region was surgically resected conservatively. [Figure 4] During surgery, there was no considerable bleeding and healed without complications. The patient has been under frequent follow-up for the past five years with no clinical indication of recurrence.

DISCUSSION

ABC is an osteolytic lesion of the head and neck that is non-neoplastic and fast in growth. We found 120 cases of ABCs of the oral cavity in our literature research. Only 13 cases of ABC of the maxilla were found,[3] and there are no cases in geriatric patients that have been reported to date when we explore in the Scopus and PubMed databases. Our case is unique because it is an ABC of the maxilla in a geriatric patient.

The etiopathogenesis and biological behaviour of ABC are still controversial. Steiner and Kantor suggested that ABCs are originated from degeneration of a pre-existing lesion like fibrous dysplasia, ossifying fibroma, giant-cell

tumour of the bone, osteosarcoma, chondroblastoma, chondromyxoid fibroma, osteoblastoma.[1] However, it is still uncertain whether the lesion is primary or after an existing bone lesion. Jaffé and Lichtenstein suggested that vascular origin because local circulatory disturbances, like malformations or dilation may increase venous pressure which may lead to bone expansion and resorption, connective tissue alteration with osteoid formation leading to cyst formation and most widely accepted theories.[2,4] Hernandez *et al*., and Panoutsakopoulos *et al*.,[5] has also been postulated that genetics may play a role, such as chromosomal translocation t (16; 17) and (q22; p13) being suggested as chromosomal disorders for the cause of main ABC.[6]

Inactive, active, or aggressive ABCs can be categorised based on clinical and radiographic features. ABC usually appears in the second or third decade of life, and there is no gender preference. The mandible was more commonly involved than the maxilla (6:1), with the posterior mandible being the most common site. It can manifest variables in clinical presentation from asymptomatic to symptomatic lesions. Painless swelling is suggested as the main symptom by some authors. Loosening or displacement of teeth, malocclusion, haemorrhage, pus discharge, nasal obstruction, lip paraesthesia and bone fracture are other associated symptoms. The presence of a dark red or brownish haemorrhagic fluid was appreciated on fine-needle aspiration is suggestive of ABC. Common differential diagnosis includes fibrous dysplasia, dentigerous cyst, radicular cyst, giant cell reparative granuloma, ameloblastoma, a simple maxillary cyst.[1]

Radiographic characteristics may be indicative of ABC, although they are not diagnostic. On orthopantomography, it appears as unilocular or multilocular radiolucency, giving the appearance of diverse patterns such as honeycomb, soap bubble, or moth-eaten due to bone disintegration with thin border.[4] A large cystic expansile mass encircled by a narrow border with internal septation was discovered on magnetic resonance imaging (MRI), indicating a hyper vascularised lesion. On a computed tomography scan, it appears usually osteolytic unilocular or multilocular radiolucent lesions.[7,8]

Histologically, ABC shows fibrous connective tissue which consists of several various size and shaped blood-filled spaces adjacent to multinucleated giant cells. There may be osteoblasts rimming around the osteoid bone, chronic inflammatory cells, extravasated erythrocytes and hemosiderin.

There are three types of ABC are described based on histopathological features, that is, solid, mixed and vascular variants.

1. Solid type: A dense stroma, scanty sinusoids, few blood vessels and caverns, bone expansion and without severe bleeding during surgery. This variant seen in the 5% of the cases.
2. Vascular variant: A loose scanty stroma, numerous engorged blood-filled sinusoids and caverns, extensive bony destruction with a spread in the soft tissues and bleeding might be encountered during surgery. This variant seen in the 95% of cases.
3. Mixed type: Combination of the two above variants.

Bony disintegration and expansion of the lesion to gingival tissue, as well as no significant bleeding, indicated that our case was of the vascular type. Because incisional biopsy alone can be difficult to evaluate, clinical and radiographic features will aid us in making the final diagnosis. The differential diagnosis of ABC based on histopathologic features includes telangiectatic osteosarcoma, giant cell tumour, reparative granuloma and brown tumour.[9]

The most appropriate mode of management for ABCs is surgical therapy, while additional options include embolization, cryotherapy, and waiting and watching. Due to the high risk of malignant transformation, such as sarcoma, radiotherapy is not recommended. The treatment of ABC varies from simple curettage to en bloc resection, depending on the age of the patient, the location and dimension of the lesion, and the extent of bone destruction.

Despite the fact that ABC is not a malignant lesion with no evidence of metastasis, there are recurrence rates following treatment. ABCs have a high recurrence rate, and they usually do so during the first year of treatment. The recurrence rate of long bones after surgical curettage and resection of ABCs was 20–50% and 11–25%, respectively, and this could be attributable to insufficient removal of the lesion.[10]

The present ABC case was rare in the anterior maxilla which was occurred in an elderly female patient. Incisional biopsy and MRI or CT scans may aid in a definitive diagnosis of the diseases with enigmatic clinical symptoms. Conservative surgical resection of the ABC is preferable with a lesser rate of recurrence due to cystic nature, bone resorption, location of the lesion and age of the patient.

CONCLUSION

The present report is the 14th ABC case describing is a rare pathologic entity of maxillary alveolus in elderly female

individuals. The main diagnostic challenge for ABC based on clinical, radiographic, and histopathologic features is the chances of misinterpreted cases. Therefore, the clinician, radiologist and pathologist need to discuss and conclude the diagnosis before the final diagnosis. Accurate diagnosis of ABC is very essential for the treatment option and to perform follow-up of the patient for assessing the recurrence.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Author's contribution

VGD wrote the paper with revision and the corresponding author.

RMB, AG put the study design, idea and collected the patients' data.

SS did the surgery and followed the patients postoperatively. All authors have read and approved the final version of the manuscript.

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Conflicts of interest

There are no conflicts of interest.

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