Aneurysmal Bone Cyst of Mandible: A Case Report

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Aneurysmal bone cysts are rare benign lesions of bone tissue, infrequent in the craniofacial skeleton about other structures like long bones or the spine. The rare jaw lesions encountered in the mandible and maxilla. On the other side, it is at the same time very exciting in terms of its differential diagnosis with other types of mandibular or maxillary bone lesions. We present the case of a 45-year-old female patient with an aneurysmatic cyst located at the left angle of the mandible describing the treatment for the same i.e. surgical excision and hemi mandibulectomy with reconstruction. We have focused on the differential diagnosis, mainly with the malignancies, which can be found at this location, along with therapeutic options classically described for these kinds of pathologies. In our patient, the surgical excision and hemi mandibulectomy allowed a complete removal of the lesion and recon plate was placed for rehabilitation.

Keywords: Ameloblastoma, Aneurysmatic bone cyst, Hemimandibulectomy, Jaw lesions

INTRODUCTION

The aneurysmal bone cyst (ABC) is one type of pseudocyst of the jaw because of lack of epithelial lining.1

ABC has been firstly recognized by Van Arsdale as an ossifying fibroma.1 Jaffe and Lichtenstein were the first to described ABC in 1942. Bernier and Bhaskar were credited with the first case report of two patients having an ABC involving the mandible in 1958.2,3

ABC is a non-neoplastic lesion of the bone, characterized by replacement with fibro-osseous tissue containing blood filled sinusoidal or cavernous spaces.4 The World Health Organization classifies ABC as a tumor-like lesion. It defines it as “an expanding osteolytic lesion consisting of blood-filled spaces of variable size separated by connective tissue septa containing trabeculae or osteoid tissue and osteoclast giant cells.”

ABCs are more commonly found in the long bones (50%) and in the vertebral column (20%). Only 1.9% of all ABCs occur in the jaws representing 1.5% of the non-odontogenic, non-epithelial cysts of the mandible.1,4

In case of craniofacial location, the mandible is more frequently affected than the maxilla a proportion from 2:1 to 11:9.2 The body and mandibular ramus are the main location with rare case reports in the coronoid process and the mandibular condyle. Age of presentation of aneurismal bone cyst is the first two decades of life, being infrequent in patients up to 20 years. There is slight sex preponderance in females.5,6

ABCs exist in two clinicopathological forms - as a primary or as a secondary lesion arising from another osseous condition - It includes fibrous dysplasia, cementifying fibroma, giant cell granuloma and certain unspecified lesions.5,7

Histologically, It Can Be of Three Variants

1. Conventional or vascular type (95%): Osteolytic lesion with blood-filled cavities and sinusoidal spaces, separated by fibrous connective tissue septa with osteoid trabeculae. Variable amount of hemosiderin and giant cells can be found

2. The solid type (5%): This form is non-cystic variant with osteoclast-like giant cells. Osteoblastic differentiation areas with osteoid and calcifying fibromyxoid tissue complete the picture7,8

3. A third form or mixed variant demonstrates features of both the vascular and solid types. It may be a transitory
phase of the lesion because sudden activation or rapid enlargement of stable lesions has been reported.⁸

This case report describes a clinically and radiographically atypical case of a large radiolucency involving the mandibular ramus. It presented as a multilocular, multilobulated lesion with irregular, but well-defined borders without an apparent sclerotic lining.

The differential diagnosis included odontogenic keratocyst, odontogenic tumor (ameloblastoma, odontogenic myxoma), giant cell granuloma, ABC and solitary bone cyst.

CASE REPORT

The case report is about a 35-year-old female who was referred to our department with the chief complaint of a hard, tender swelling in the left lower back teeth region since 1 month. She gave a history of a small swelling that had started 3 months back and had since then increased in size with associated pain for the past 1 month. Medical records and family history were unremarkable, and there was no history of trauma.

Clinical examination was normal except for apparent facial disharmony with a firm, smooth, tender, diffuse swelling of 5 cm² × 5 cm² in the left mandibular body, angle and preauricular region.

Intra orally, the expansion was evident in both the lingual and buccal vestibular areas with intact mucosal tissues. There was no sinus opening or any discharge. Teeth in the involved areas were vital as checked by both thermal and electrical pulp testing.

Radiographical examination showed a uni-locular radiolucency extending from the root of the right second premolar to the distal aspect of the third molar up to sub sigmoid region (Figure 1). Resorption of roots of the involved teeth was not observed. There was no discontinuity of the lower border. Routine hematological investigations were non-contributory. On aspiration, a volume of 2 ml of blood was obtained.

Considering the clinical presentation, radiographic features and positive blood aspiration, a provisional diagnosis of ABC was made, and surgical treatment for resection of the mandible, followed by immediate reconstruction with a titanium recon plate was planned. Under general anesthesia, through an extra oral submandibular approach the tumor was exposed (Figures 2 and 3). The mandible was resected 1 cm beyond the radiographic margins both at the ramus and the premolar-molar regions. The jaw cortex was ballooned out and thinned having an eggshell appearance, but without loss of continuity.

There was infiltration into the soft tissues in areas of perforation of the buccal cortex, which was cleared (Figure 4).

Primary reconstruction of the defect was carried out with a titanium reconstruction plate (Figure 5). There was no significant blood loss while surgery and closure were completed using staples (Figure 6). Patient was put on intermaxillary fixation for 3 weeks.
DISCUSSION

The ABC, as a distinct pathologic entity, was first described in the literature by Jaffe’ and Lichenstein.²,⁹ The term “aneurysmatic” refers to the “blow-out” effect or expansion of the affected bone that appears in this type of lesions.

The long bones and vertebrae are more commonly affected than facial bones. They are rare in the jaws, and the mandible is affected twice as frequently as the maxilla.⁴,⁵,⁸ The molar region, the angle and the ascending ramus of the mandible are the most affected sites.⁶,⁸ The lesion generally affects young persons predominantly under 20 years of age, with a slight predilection for females.¹₀

Clinically, there is a firm, non-tender, or slightly tender swelling for a period of 1 week-3 years which enlarges progressively expanding and occasionally perforating the bony cortex, displacing the teeth, which, however, remain vital.⁴,⁸,¹⁰ Sometimes, a history of rapid growth is elicited, possibly as a result of the erosion of the cortical plate.⁴ The overlying mucosa remains normal.¹⁰

There are typical roentgenographic findings: The bone cortex can also be expanded. The multilocular effect gives this cyst the characteristic but no pathognomical “honeycomb” and “soap bubble”-like appearance seen in other lesions such as giant cell granuloma, myxoma, desmoplastic fibroma, haemangioma, keratocyst, ameloblastoma and other tumors. Occasionally, destruction of the bony cortex may be identified, displaying a periosteal reaction imaging or “ray-sun” effect that is characteristic of osteosarcomas.⁴,⁸,¹¹,¹²

Lesions are radiolucent in most cases but can be radiopaque or mixed. Computerized tomographic scans may show a uni-cystic or multilocular lesions with cortical expansion, destruction or calcification along with a circumscribed
subperiosteal reaction or a moth-eaten appearance. Diagnosis based solely on the radiographic appearance is impossible, because there are other entities with similar radiographic appearance, such as ameloblastoma, myxoma, central giant cell granuloma, odontogenic cysts, or central hemangioma of the bone.

The contents are similar to the reddish brown material found in giant cell lesions. In addition, there is a persistent, slow swelling of blood from the lesion during surgery. Histologically the ABC consists of many sinusoidal blood-filled spaces set in a fibrous stroma. Multinucleated giant cells are patchily distributed in areas of old hemorrhage, and there is often evidence of osteoid and bone formation and hemosiderin is present in variable amounts. The cause of these lesions is still controversial. Jaffe and Lichtenstein refer to altered hemodynamic state leading to a dilated congested vascular bed, causing resorption and erosion of the bone with connective tissue replacement and osteoid formation.

Struthers and Shear, who postulated that ABCs are secondary lesions related to degeneration of a pre-existing bone lesion like the central giant cell granuloma, fibrous dysplasia, ossifying and cementifying fibromas. Some pathologists assume the ABC is a response to an alteration in the vessels of the region causing a proliferative reaction without healing owing to continual effusion of blood from the injured vessels.

The lesion has also been found in association with fractures and other bone injuries. Although, the clinical appearance may be an aid to proper diagnosis, a definitive determination can be made only after histological examination of the excised specimen.

Surgical curettage or excision is the treatment of choice and hemorrhage is not, in fact an operative problem. In an attempt to reduce the potential for recurrence, surgical resection of the affected bone has been employed. Radiation was used; however, this implies a risk for the appearance of a radiation sarcoma.

The findings of this patient corresponded with those of Struthers and Shear. This along with the sudden growth, bilateral expansion, cortical destruction, osteoid formation and tumor-like appearance can easily cause confusion with malignancy. Immediate mandible reconstruction using a titanium recon plate is recommended.

CONCLUSION

Concerning the treatment, although many options have been performed, the gold standard is still the surgical excision and curettage of the cavity. The use of radiotherapy is not recommended because of the probability of radio induced tumors.

REFERENCES


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