Case Report

Giant aneurysmal bone cyst of the mandible: A case report and review of literature

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ABSTRACT

Aneurysmal bone cysts are rare benign lesions of bone tissue. They are composed of vascular spaces blood-filled and surrounded by fibrous tissue septa. They are considered as pseudo cysts because of lack of epithelial lining. Here, we describe a giant case of ABC in 12-year-old female child having a massive swelling over the right side of the mandible treated with segmental resection and reconstruction with a reconstruction plate. Case is also discussed with the review of literature.

Key words: Aneurysmal bone cysts, benign bone lesions, mandible, pseudocysts

INTRODUCTION

Jaffé and Lichtenstein were first to recognize ABC affecting the metaphyseal region of long bones. Berneir and Bhaskar described the first case of ABC in 1958.[1,2] Fifty percent of ABC arise in long bones 20% in vertebral column and 1.5% in mandible.[3,4] It affects young person under 20 year of age with no gender predilection.[4,5]

It can be classified into three types. Conventional or vascular types manifests as a rapidly growing expansive, destructive lesion causing cortical perforation and soft tissue invasion. The solid type may present as a small asymptomatic lesion first noticed as a small asymptomatic lesion on a routine radiograph.[6,7] 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.[7]

These are nonneoplastic but locally aggressive tumor with occasional rapid growth, which should be differentiated from other multilocular processes like ameloblastoma, ossifying fibroma, epithelial cyst, giant cell granuloma.[1]

CASE REPORT

A huge mandibular swelling six months before started as a small swelling and gradually progressed to present size, her medical and family history was remarkable. On extraoral examination there was facial asymmetry with apparent massive swelling involving the right side of the lower jaw measuring approx. 6.8 * 7.2 cm. The swelling was firm and non tender [Figure 1].

Intra oral examination reveals a diffuse swelling in relation to the right lower first premolar, second premolar and first and second molar with displacement of first and second molar, there was cortical plate expansion and erosion on the both the sides with huge soft tissue mass obliterating the buccal, as well as the lingual sulcus pushing the tongue to the contralateral side. The superior surface of the mass was in the contact with maxillary posterior teeth, which were causing ulceration on the superior surface of the mass [Figure 2].

The child had a very poor oral hygiene with drooling of the saliva and difficulty in mastication, on aspiration rapid filling of the syringe with frank blood took place there was no bruit on auscultation.

A panoramic radiograph [Figure 3] revealed a large
multilocular radiolucency involving the right body of mandible extending from distal of first molar and extending on to the ramus involving the inferior border of the mandible completely, and going up to the coronoid process and subcondylar region.

MRI of the lesion revealed a large lobulated expansile mass with central necrotic hemorrhagic areas measuring 6.8 * 7.7 * 6.8 cm in anteroposterior, transverse and superoinferior dimension. Multiple vascular flow voids in the lesion arising from right ECA, lingual and facial artery [Figure 4].

In view of the large size of the lesion, treatment plan of segmental resection was made, after ensuring patient fitness for GA, the patient was taken for segmental resection under fiber optic NET intubation, an extended neck crease incision was given and the lesion was exposed in its entirety ECA was dissected out and a loop was kept ready to be used in case of extensive hemorrhage.

The mass was excised with safe margin and was reconstructed with Titanium Angled Reconstruction plate.

Gross examination of the mass revealed the cortical expansion with erosion and replacement of endosteal tissue with large dilated blood filled spaces with tissue necrosis and evidence of incompletely osteoid tissue. There was no significant blood loss after surgery.

The tissue was send for histopathological examination, microscopic examination reveals large lined by endothelial cells and there was no presence of giant cells [Figure 5].

Patient made an uneventful recovery [Figure 6].

Reconstruction plate was well contoured with the mandible [Figure 7].

**Discussion**

The term aneurysmal is a misnomer and refers to the
blow out effect or expansion of the affected bone that appears in this type of lesions.\[8\]

It is a pseudo cyst lacking epithelial lining,\[1,5,8\] it comprises the 5% of all the lesion of craniofacial and maxillofacial bones.\[2\]

The etiopathogenesis of ABC is highly debatable and controversial and many theories have explained their origin, Jaffé and Lichtenstein have suggested a vascular origin in which there is alteration in local hemodynamics causing increased venous pressure and engorgement of the vascular bed leading to the resorption.\[1,9\]

Steiner and Kantor have suggested that ABC can develop as primary or secondary lesion associated with other bone diseases, Struthers and Shear have also concluded that ABC can occur as a secondary phenomenon in a preexisting lesion and giant cell granuloma.\[3\]

Levy et al., has proposed a history of trauma and subperiosteal hematoma formation is a essential factor in the development of ABC, however, Tillman et al., have reported 95 cases with no history of trauma.\[7\]

Hernandez et al., classified ABC as primary and secondary, primary could be congenital or acquired. The congenital is seen in children and young adults with no history of trauma whereas the acquired is found in adults with history of trauma. The secondary is postulated to be associated with degeneration of preexisting lesion such as cyst, tumor or fibro osseous lesions.\[9\]

The two lesion can exist independently, hence, ABC is considered as non neoplastic fibro‑dysplastic, non cystic bone entity.\[3,10\]

In our case also there was no history of trauma so etiology could be either due to alteration in local hemodynamics or degeneration of any preexisting lesion at the involved site.

Panoutsakopoulos et al., had described three cases with chromosomal anomalies involving band 16q22.

Familial incidence has been reported in the literature.\[11‑13\]

ABC are most commonly found in long bones and in vertebral column 1.9% reported in jaws and unusual location like condyle and coronoid process been reported.\[3,7,8,14\]

It is extremely variable in clinical presentation ranging from small lesion to rapidly growing expansible destructive lesion causing deformity, pain and cortical perforation.

The latter presentation was found in our case also where there was rapidly expansile growth with the short history.

The radiological features are quite conflicting, it may appear radiolucent, radio‑opaque or mixed. The bone can appear expanded, cystic resembling a honey comb or soap bubble in appearance similar to what is seen in benign odontogenic tumors and diagnosis. Based on radiographic appearance impossible because there are multiple lesions having similar radiographic appearance such as Ameloblastoma, Myxoma, Central giant cell tumor and central hemangiomas.\[15\]

Our lesion also had a multilocular appearance resembling other odontogenic lesions.

**MRI features**

MRI is mandatory in complex cases such as the reported case in order to improve the plain radiographic examination and CT scans as it is more accurate in soft tissue contrast.
MRI finding of fluid filled levels inside the lesion are highly specific of ABC.\[16\]

In our case also MRI clearly depicted the flow voids arising from right ECA, lingual and facial arteries.

Angiography is sometimes used when MRI shows hypervascularization.\[8\]

Histologically, ABC consists of blood filled spaces separated by fibrous septa with multinucleated giant cells and osteoid. Hemosiderin is present in variable amount and there is evidence of osteoid and bone formation, this is characteristic of classic or vascular form.

The histopathological feature in our case was consistent with the above mentioned features, however, we did not find presence of any giant cells in our lesion.

Solid form is the other type which is a non-cystic variant with solid grey white tissue with osteoid and calcifying fibro-myxoid tissue. The mixed form demonstrates elements of both vascular and solid type.\[8\]

Treatment usually directs to complete removal of the lesion, the modalities are enucleation and curettage. Diagnostic and therapeutic embolization, segmental resection and reconstruction. Simple curettage is associated with high recurrence but few authors have reported no recurrence following simple curettage of mandibular lesion.\[1,17\]

Segmental resection must be done in cases of multiple recurrence or extension to the overlying tissue.\[1,8,14\]

Recurrence rate ranges from 20-30% and seem to occur more frequently, first year after surgery.\[19\]

Self healing cases have also been reported on long term follow-up.\[19\]

Several author recommends immediate reconstruction of the defect with autografts in cases of aesthetic deformity high-risk of fracture and loss of mandibular continuity.\[3,5,14\]

The present case was treated by segmental resection and reconstruction by a reconstruction plate due to the size and the extension of the lesion reconstruction, with free tissue transfer was planned for a later date.

**CONCLUSION**

A case of giant ABC of mandible in a 12-year-old female child treated segmental resection and reconstruction of the defect with a reconstruction plate, is presented along with review of literature.

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