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Aneurysmal Bone Cyst Secondary to Ossifying Fibroma- Case report and review of literature

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Abstract: *Aneurysmal bone cyst is an uncommon non- neoplastic osteolytic bone lesion that was first described by Jaffe-Lichtenstein in 1942, whichless frequently occurs in maxilla. Various theories support the origin and differentiate it as primary and secondary lesion. We represent a case of 11 year old girl with a bony hard swelling on left maxilla with a multilocular radiolucent andhighly vascularized lesion with thin internal septation present. Treatment includes conservative surgical excision of mass.Histopathological finding supported the diagnosis of aneurysmal bone cyst as a secondary lesion in association with ossifying fibroma.*

Keywords:*aneurysmal bone cyst, giant cells,maxilla, ossifying fibroma*

I. Introduction

Aneurysmal bone cyst (ABC) is non-neoplastic benign osteolytic lesions which less frequently occur in head and neck.[1] According to literature, 66% occur in jaw with mandible considered as most frequently affected site than maxilla.[2,3,4] According to WHO, ABC is considered as a benign intra-osseous lesion characterized by blood filled spaces of varying size associated with fibroblastic stroma containing multinucleated giant cell, osteoid, and woven bone.[7,8]There are various theories that support the origin and differentiate it as primary and secondary lesion. Theories range from post-traumatic, reactive vascular malformation to genetically predisposed bone tumor.[9] ABC is considered as reactive entities that are either related to circulatory disorder causing increased venous pressure or to a local vascular disorder attributable to preexisting lesion.[10] Other theory could be a congenital primary lesion that may coexist with other osseous pathologies. Another pathological mechanism could be bone tumor that may facilitate these hemodynamic alterations. Other different theory proposed that aneurysmal bone cyst is a tumor rather than reactive lesion and are secondarily related to degeneration of pre-existing bone lesion such as central giant cell granuloma, fibrous dysplasia or ossifying and cementifying fibroma which can be confirmed by histopathological examination. [11] Recent studies done on primary ABC proposed the primary lesion as a tumor rather than a reactive lesion. [9]
Despite of controversy done on its origin by various authors, clinical presentation of the lesion is non-specific andcan further lead to misdiagnosis as a highly-aggressive or malignant neoplasm by its presentation as a rapid expansive locally destructive growing mass. A definitive clinical diagnosis and histopathological examination can lead to correct diagnosis of this entity. [11]

To best of our knowledge, in past 58 years only 40 cases was reported in maxilla with a single case report associated with ABCsecondary to ossifying fibroma. We report an additional case of ABC of maxilla associated with ossifying fibroma considering it as a 41th case to be added in literature with involvement of secondary lesion and confirming the theory proposed with a brief literature review.

II. Case Report

An 11 year old girl reported to the department of Oral Medicine and Radiology for evaluation of rapidly growing mass on left side of maxilla since 3 months (fig.1a). Associated symptoms were non-contributary. On inspection, swelling was seen on left malar region with normal overlying skin (fig. 1b). The swelling was bony hard and non- tender. Intraorally, vestibular swelling was obvious in 23,24,25,26 regions with expansion of buccal cortical plate. There was root stump in relation to 65 (figure 1c). Further, the radiographic investigation revealed a unilocular radiolucent lesion involving the alveolar ridge from 24, 25, 26 and unerupted 27 (fig 2). Anosteolytic lesion was seen in cone-beam computed tomography involving whole of left maxilla(fig 3a). Coronal, axial and tangential sections showed expansile lesion displacing the maxillary sinus, nasomeatal complex and inferior orbital rim size ranged 31.47mmantero-posteriorly, 47.23mm medio-laterally and 32.73mm super-inferiorly.The periphery of lesion had interrupted corticated margins. Internal

structure showed hazy multilocular appearance with surrounded wispy septa emanating at right angle from periphery of lesion(fig 3b andc).

Aspiration showed dark red hemorrhagic content.Presuming a highly vascularized tumor, CT angiography was carried out which showed areas of hyperdensity in non-contrast study suggestive of hemorrhagic content. No arterial feeder was found, venous phase showed heterogenous enhancement and fluid density areas(fig 4a, b,c). Looking into the clinical and radiological features, we considered it as central giant cell granuloma.

Before going for treatment, blood chemistry and hematologic profile was carried out in which serum alkaline phosphatase levels were elevated (453U/L). Surgical treatment with conservative resection and immediate reconstruction of the defect was performed (fig 5).

The histopathological examination of surgical sample was carried out in which the tissue was divided into four parts first the pathological part showing sinus lining with proliferative cystic growth which was grayish in color, second part was the maxillary sinus lining and third and fourth part was curetted tissue. Upon hematoxylin and eosin staining, First part showed cavernous spaces (not lined by epithelium) of varying size, filled with erythrocytes surrounded by cellular, fibroblastic stroma containing numerous multinucleated giant cells, bony trabeculae and woven bone suggesting the diagnosis of aneurysmal bone cyst. The third and the fourth part showed cellular fibrous connective tissue stroma with numerous spherules of calcified material and bony trabeculae with osteocytes in lacunae and osteoblastic rimming suggesting the features of ossifying fibroma (figure 6a&b).

The final diagnosis after histological examination was aneurysmal bone cyst secondarily associated with ossifying fibroma. There were no signs of recurrence seen after 7 months of recall visit.

III. Review of Literature

Literature search of reported cases of ABC occurring in jaw for the 58 years revealed 104 cases occurring in both maxilla and mandible. According to literature 40 cases are reported in maxilla and 64 cases in mandible. The reported cases of maxilla are summarized below in Table- 1.

According to review of literature done the clinical findings suggest that mandible is more commonly affected than maxilla with maximum reported cases. The clinical findings suggest that ratio of occurrence in female is more than in male with a ratio of 2:1. The age presentation of its appearance is more in younger individual but it may be bimodal. The peak incidence of its appearance is in first and second decade of life but some cases are also seen in elderly group. The etiology associated with lesion is less associated with trauma and more towards unknown origin. The radiological findings reported are unilocular and multilocular with more radiolucent than radiopaque. According to theories and reviews done ABC is more a primary lesion than a secondary lesion associated with any other osseous condition. Presently, reported cases of secondary lesion in mandible are four and in maxilla are eight with only 1 case associated with ossifying fibroma in maxilla. The treatment of choice in most of the cases is curettage with less of resection accompanied with bone graft. The recurrence rate is less with a follow up period ranging from 6 month to maximum of 35 years.

IV. Discussion

Aneurysmal bone cyst was first described in literature by Jaffe et al. [12]The term “aneurysmatic” refers to the “blow-out” effect or expansion of the affected bone that appears in this type of lesion.[13]Age of presentation is generally first two decades of life with mandible being the most commonest site than maxilla. Sex predilection is more in female than male. The clinical presentation is non-specific can present as a rapid growing mass that can be misdiagnosed as a highly aggressive tumor or malignant neoplasm. [11,13]The lesion being locally aggressive can be differentiated with other multicystic lesion like ameloblastoma,ossifying fibroma, and giant cell granuloma.[2,13]The classical clinical description of aneurysmal bone cyst is well defined swelling of soft tissue due to expansion of adjacent bone causing facial asymmetry. There is slow progressive growth until cortical plates are eroded and then rapid growth. [11] Pain is associated but not considered as significant features of ABCs. In our case the patient supports the clinical description. Radiological examination shows presence of cystic radiolucent imaging, usually multilocular, with a cystic meshwork divided by septa. [11]According to literature, the lesion is described as an eccentrically loculated, ballooned-out, multilocular radiolucency with a honey-comb or soap bubble appearance. Other findings are dense filamentous septa converging on central of lesion with thinning and destruction of the cortex well appreciated in CT scan or CBCT.[52] This type of appearance can be similar to other lesion of jaw such as Giant cell granuloma, Myxoma, Desmoplastic fibroma, Hemangioma or Ameloblastoma. Angiography is only done when hemangioma or high grade vascularization is suspected. [11]Other finding such as biochemical test showed increase in serum alkaline phosphatase supporting lesion as fibro-osseous in nature. FNAC revealed the lesion as a highly vascularized with appearance of dark red hemorrhagic content. The appearance of two type of behavior in one lesion can be misdiagnosed as a hybrid lesion as they are the lesions consisting of association of

features from different pathologies. Controversy regarding the nature of lesion was confirmed by histopathological analysis which showed the lesion as a fibrous connective tissue stroma containing many cavernous or sinusoidal blood-filled spaces. Presence of many fibroblast as well as multinucleated giant cell and hemosiderin found in these type of lesion. [9,11,52]The histopathological analysis confirmed that ABC is secondarily associated with a fibro-osseous lesion.

Histopathological analysis of present case where signs of other pre-existing lesion were found supported the theory that ABC are secondary lesion related to degeneration of pre-existing bone lesion.[11] Review of literature supports that ABC can be associated with primary as well as secondary lesion (as shown in table-1) and is presumably more a reactive lesion with two clinicopathological entity.

Nowadays treatments modalities of this lesion are highly controversial with high recurrence rate. It includes surgical resection of the lesion with immediate reconstruction of the defect with autogenous graft in case of aesthetic deformity. Curettage can also be carried out with careful follow up. Supplement procedures include cautery,cryotherapy, and radiation therapy but are considered to be unsuccessful and result into sarcomatous change. Long term follow- up is required considering it as a highly reactive lesion in association with other osseouscondition.

V. Conclusion

Aneurysmal bone cyst shows marked variability in clinical and radiological diagnosis. Although various theories and pathogenesis support that it is a tumor but ABC could be considered as a reactive lesion having two distinct clinicopathological forms either primary or secondary and we as a radiologist should considered it as distinct entity in our practice. Hence, considering the recurring rate of lesion, close clinical and radiological follow up is recommended.

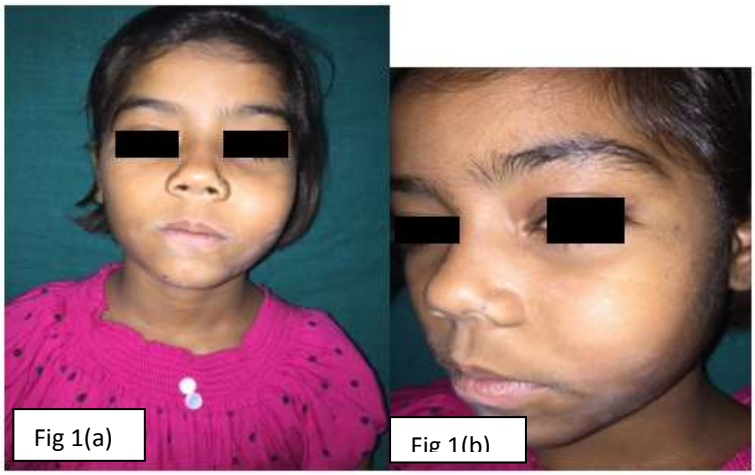


Fig 1(a and b) clinical photograph showing swelling involving left side malar region with normal overlying skin

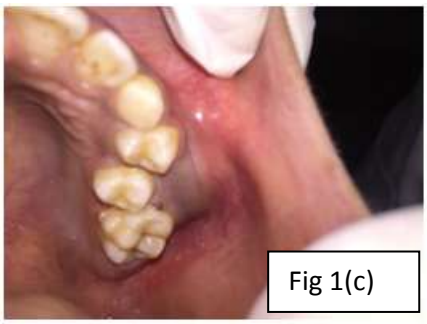


Fig 1(c) intraoral photograph showing vestibular swelling with buccal cortical plate expansion and root stump in relation to 65

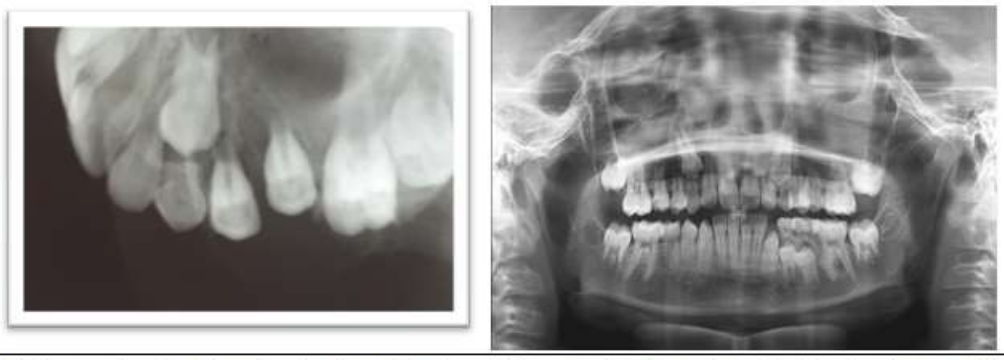
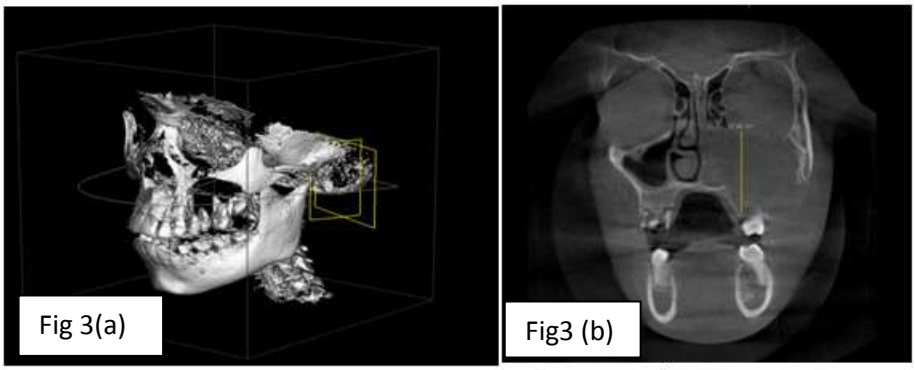


Fig 2 conventional radiographs showing unilocular radiolucent lesion in relation to 24, 25, 26 and unerupted27



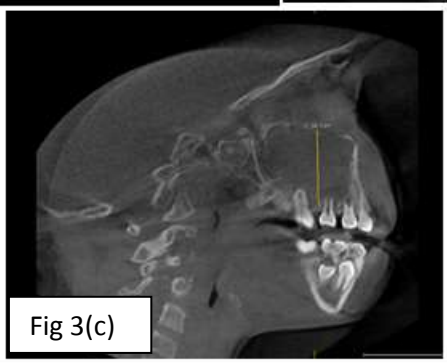


Fig 3(a) CBCT showing a destructive lytic lesion Fig (b & c) coronal, sagittal and axial section showing interrupted cortical margin with thin internal wispy septa emanating at right angle from periphery.

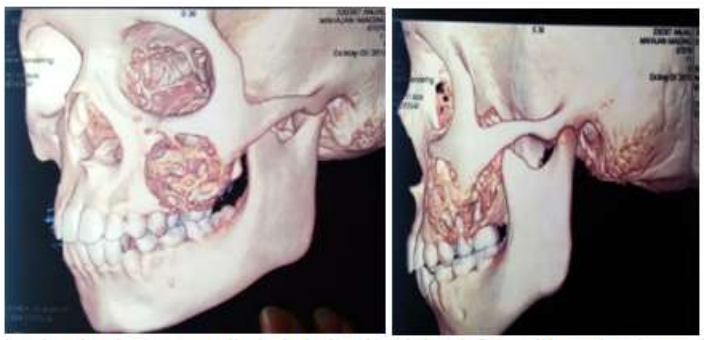


Fig 4a: showing large expansile lytic lesion involving left maxillary alveolar process.

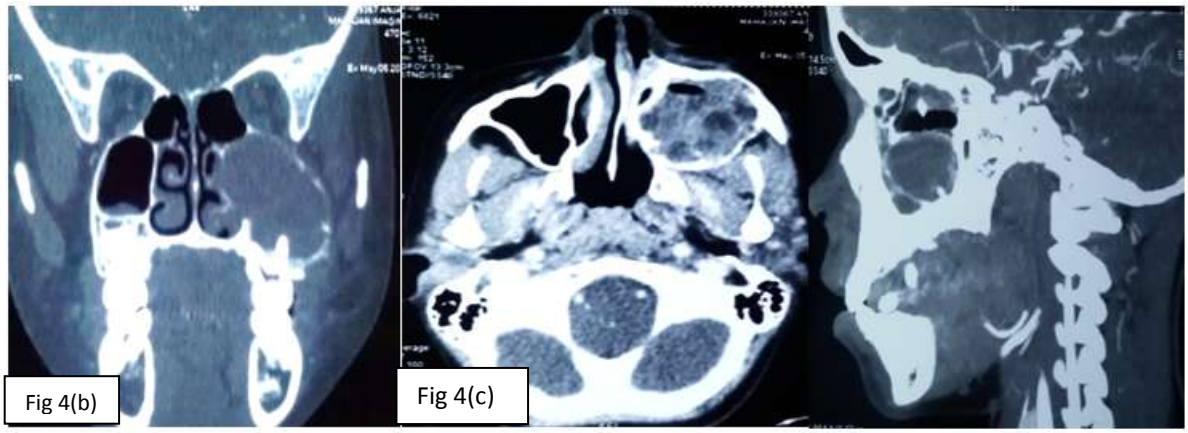


Fig 4b&c: CT angiography showing areas of hyperdensity in non-contrast study suggestive of hemorrhagic content and multiple areas of fluid density.

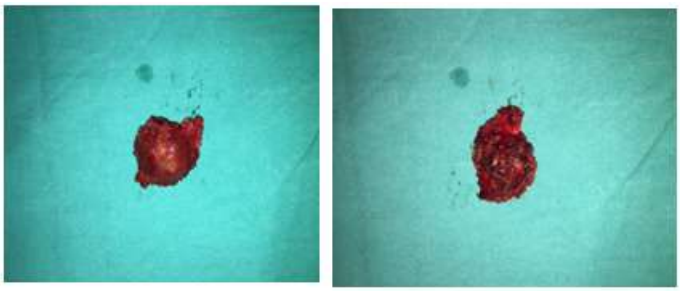


Fig 5: gross picture of resected cystic lining

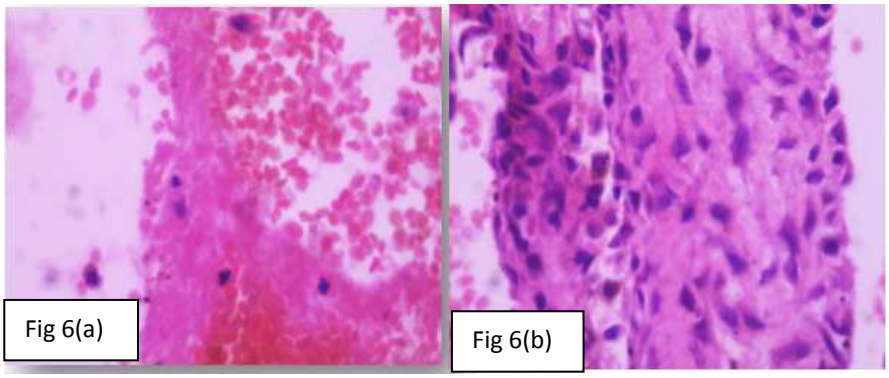


Fig s6a&b: photomicrograph(H&E,X40) of the section showing cellular fibroblastic stroma with numerous multinucleated giant cell, bony trabeculae and woven bone.

Table 1- Recorded Aneurysmal Bone Cyst Of Maxilla

AUTHOR	AGE (YEARS)	SEX	RADIOLOGICAL FEATURES	DETAIL OF LESION
Bhaskar et al (1959)[14]	22 yrs	Female	Radiolucent	Primary
Wang et al (1960)[15]	8 yrs	Female	Multilocular radiolucent	Primary
Vianna and Horizonte (1962)[16]	18yrs	Male	Multilocular	Primary
Yarington et al (1964)[17]	48 years	Female	NA	Secondary to giant cell granuloma
Gruskin&Dablin (1968)[18]	20 yrs	Male	NA	Primary
Nosaka&Nakazawa (1968)[19]	8yrs	Male	NA	Primary
Byrd et al (1969)[20]	17 yrs	Female	Multilocular	Primary
Lejeme&Bordelon (1970)[21]	17 yrs	Female	NA	Primary
Ellis & Walter (1972)[22]	17 yrs	Male	Radiopaque	Secondaryto cementifying fibroma
Romaniuk& Becker (1972)[23]	10 yrs	Male	Multilocular radiolucent	Primary
Komorn et al (1972)[24]	26 yrs	Female	NA	Primary

Power & Glaser (1975)[25]	16 yrs	Female	Radiolucent	Primary
Roa et al (1975)[26]	12 yrs	Male	NA	Primary
Ruiter et al (1977)[27]	NA	NA	NA	NA
Coryn et al[28]	9 yrs	Male	NA	Secondary to fibrous dysplasia
Reyneke (1978)[29]	18 yrs	Male	Unilateral radiolucent	Primary
Boyd et al (1979)[30]	27 yrs	Male	Radiolucent	Secondary to ossifying fibroma
Kaine et al (1979)[31]	14 yrs	Male	NA	Primary
Salmo et al (1981)[32]	55 yrs	Male	Unilateral radiolucent	Primary
Robinson et al (1985)[33]	13 yrs	Male	Multilocular radiolucent	Secondary to cementifying fibroma
Zacharaides et al (1986)[34]	35 yrs	Female	Radiolucent	Primary
Takimoto et al (1990)[35]	15yrs	Female	NA	Secondary to fibrous dysplasia
Hady et al (1990)[36]	13 yrs	Female	Multilocular radiolucent	Primary
Hardy et al (1992)[37]	29 yrs	Male	Unilateral radiolucent	Primary
Matt et al (1993)[3]	12 yrs	Female	NA	Primary
Medeiros et al (1993)[38]	19 yrs	Female	Multilocular	Primary
Cohen et al (1993)[39]	13 yrs	Male	NA	Primary
Cheynet et al (1994)[40]	14 yrs	Female	NA	Primary
Wojno&McCarthy (1994) [41]	22yrs	Female	NA	Secondary to cementifying fibroma
Suzuki et al (2001)[42]	23 yrs	Male	Multilocular radiolucent	Primary
Pasquini et al (2002)[43]	5 yrs	-	NA	Primary
Yasuoka et al (2002)[43]	16 yrs	Female	Radiolucent	Secondary to central giant cell granuloma
Weir et al (2003)[44]	10 yrs	Female	NA	Primary
Sanchez et al (2004)[45]	14 yrs	Female	Multilocular	Primary
Guzman et al(2005)[46]	15yrs	Female	Multilocular radiolucent	Primary
Fyrmpas et al (2006)[47]	12 yrs	Female	Multilocular	Primary
Saheeb et al (2007)[48]	5 yrs	Female	Radiolucent	Secondary to cementifying fibroma
Tang et al(2009)[49]	17 yrs	Male	Multilocular	Primary
Mohammed et al (2015)[50]	15 yrs	Male	Radiolucent	Primary
Debnath et al (2016)[51]	8 yrs	Male	Multilocular	Primary

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