AMELOBLASTIC FIBROMA: A RARE CASE REPORT
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ABSTRACT:
Ameloblastic fibroma is an extremely rare mixed odontogenic benign tumor. This tumor is commonly seen in young adults and clinically manifested as a slow-growing swelling associated with impacted tooth. This report describes voluminous ameloblastic fibroma in 45 year old male in the mandible and is not associated with impacted which is very rare presentation. The radiograph revealed a well-defined multilocular lesion in the mandible. The lesion was excised and reconstructed with titanium plates. To conclude, though Ameloblastic fibroma is a rare neoplasm should be considered as differential diagnosis, irrespective of age and its association between impacted tooth.

Key words: Odontogenic tumor, Ameloblasticfibroma(AF), multilocular lesion, impacted tooth.

INTRODUCTION
Ameloblastic fibroma (AF) is also known as fibrous adamentanoma, soft mixed odontogenic tumor, soft mixed odontoma, fibroadamentoblastoma. AF is an extremely rare mixed odontogenic benign tumor of odontogenic origin comprising about 1.5-4.5% of all odontogenic tumors.[1] It usually occurs in young patients being diagnosed at a mean age of 15 years. Males show slightly higher prevalence (M: F = 1.4: 1), and the posterior mandible is the most common anatomic location. Diagnosis is generally made through routine radiographic examinations performed to look for an impacted tooth as a cause of the swelling.

In 1946, Thoma and Goldman were the first to classify this tumour as a separate entity. It has both the epithelial and ectomesenchymal elements are neoplastic. This group of lesions are referred to as mixed odontogenic tumors that histological resemble various stages of tooth formation. AF is defined by WHO as “consisting of odontogenic ectomesenchyme resembling the dental papilla and epithelial strands and nests resembling dental lamina and enamel organ.

It is commonly manifestation as a slow-growing swelling. Most of the cases are asymptomatic and discovered during routine oral examination as a unilocular radiolucent lesion. This rare enlarging lesion usually has a multilocular radiolucent pattern. The neoplastic nature of AF is often suggested by the fact that some of these tumors could recur following surgery.[2] These cases should be followed up as there are few reported cases of malignant transformation from a pre-existing AF.[3]
Here is a reported case of AF with a rare presentation.

**CASE DETAIL:**

A 45 year old male patient with the chief complaint of swelling and facial asymmetry on the left side since 2 years visited outpatient department with a chief complaint of discomfort mastication and speech. He reported with no pre-existing medical conditions, fever, or other signs of infection. A physical examination revealed facial asymmetry extra oral swelling involving lower border of mandible with an increase in volume extending from the left body of the mandible (7 cm)(figure 1). Intraorally, an expansile mass of left of the buccal vestibule extending posteriorly 37 anteriorly to distal surface of 33(figure 2). It was a rubbery, in consistency non-tender with poorly defined boundaries and a smooth surface. This lesion was associated with a obliteration of vestibule and tooth mobility of associated teeth. Medical and dental histories were non contributory.

Panoramic radiograph revealed a multilocular radiolucent lesion of left mandibular body extending posteriorly from distal root of 37anteriorly crossing the midline extending to 41 (figure 3).

Occlusal radiograph showed expansion of both the buccal and lingual cortical plates (figure 4).

The CT panoramic findings revealed soft heterogeneous predominantly hyperdense tissue attenuation extending in body of mandible on left side causing fenestration of the same(figure 5).3D reconstruction indicated destruction of the cortical bone involving both buccal and lingual cortical plates causing fenestration (figure 6).

Fine needle aspiration showed negative result. Incision biopsy was performed.

Macroscopically, whitish soft tissue of rubbery texture was observed.

Biopsy specimen was taken for histopathological examination which showed strands, cords and islands of odontogenic epithelium in a primitive connective tissue stroma. The epithelial islands and cords were characterized by peripheral columnar hyperchromatic cells and were frequently only two cell layers thick (figure 7). The mesenchymal component consisted of evenly distributed plump ovoid and stellate cells in a loose myxoid to predominantly eosinophilic matrix resembling the primitive dental papilla. No hard tissue structures were detected. These findings are suggestive of AF (figure 8). Based on the clinical features, radiological imaging, and histopathological reports, a diagnosis of AF was established.

Mandibular resection, from left posterior ramus to contralateral canine, was performed with primary reconstruction using titanium reconstruction plate. Post operative intra orally examination after 1 month showed signs of healing (figure 9).

Post operative panoramic radiograph after 1 month shows healing of the bone (figure 10).The patient was followed up
for 1 years; the outcome was functionally and aesthetically satisfactory and there was no signs of recurrence.

**DISCUSSION**

AF is a true-mixed neoplasm of odontogenic origin with both epithelial and mesenchymal tissues.[4] These neoplasms are noticed in young patients especially in the first two decades of life and mandible is considered to be the most common site of occurrence than the maxilla by a factor 3.1. In contrast to this, the present case showed lesion in an adult patient in the fifth decade of life. Males are more commonly affected than females, who are usually diagnosed between the first and second decades of life frequently presenting with a painless swelling of the jaw. AF is diagnosed between the first and second decades of life.[6]

It is hard to say that delay in seeking treatment was the sole reason for the late diagnosis as the patient had had symptoms for only 2 years and it would be difficult to prove the lesion was present but asymptomatic for over 30 years until the late diagnosis in this case.

AF is generally associated with enclosed teeth in the posterior region of the mandible, angle or ramus but in present case lesion was not associated with impacted tooth, but was associated with missing first molar. Bone expansion and tooth dislocation are common findings, similar finding were present in this case.

The patients usually present with a asymptomatic firm to hard swelling, but intraoral ulceration, or tenderness may also be observed, present case also showed similar presentation but without any intraoral ulceration or tenderness.

Radiographically they appear unilocular or multilocular with smooth well-demarcated borders.[7]

Cortical expansion of the affected bone is commonly observed which was noted in the present case elucidating its true neoplastic nature.

This case reported also showed huge multilocular radiolucency with indistinct curved septa. In the case reported, revealed root resorption and with no enclosed teeth; these are unusual findings for this kind of lesion. The tomography images revealed fenestration of the cortical bone, a characteristic of extensive and long-standing lesions but which is rarely observation in AF.

Microscopically the epithelial component occupies the mesenchymal stroma in various patterns like thin long strands, cords, nests, or islands. Unlike the strands in ameloblastoma, the strands in AF exhibit double or triple layer of cuboidal cells The ectomesenchymal component is composed of typical plump fibroblasts with delicate collagen fibrils simulating the dental papilla.[9]

The following lesions should be considered be considered under differential diagnosis.(table 1)
An extensive surgical treatment is suggested as the initial approach due to its high recurrence rate (18%) and the greater chances of recurrent AFs transforming into ameloblastic fibrosarcoma (45%).[9]

Patients with AF must be followed up for a long period to enable the early detection of possible recurrence or malignant transformation. This reported case is followed up for 3 years, no recurrence or malignant transformation is detected and still is under follow up.

CONCLUSION:
A careful treatment planning is necessary considering their recurrence rate and ability to undergo malignant transformation. Though Ameloblastic fibroma is an rare neoplasm should be considered as differential diagnosis, irrespective of age and its association between impacted tooth.

REFERENCES:


FIGURES:

Figure-1: Voluminous swelling on left side of mandible

Figure-2: Intraoral picture showing obliteration of left buccal vestibule.

Figure-3: OPG showing extensive multilocular lesion on left side of the mandible.

Figure-4: Occlusal radiograph showing expansion of lingual and buccal cortical plates.

Figure-5: CT Panoramic view

Figure-6: 3D reconstruction.
Figure-7: Histopathogical picture showing odontogenic epithelium in connective tissue.

Figure-8: Histopathogical (100X) showing stellate cells in a loose myxoid resembling the dental papilla.

Figure-9: Post operative picture after 1 month.

Figure-10: Postoperative OPG after 1 month