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CASE REPORT

CLINICOPATHOLOGICAL ASPECTS OF AMELOBLASTIC FIBROMA- A CASE REPORT

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ABSTRACT

Ameloblastic fibroma is an uncommon benign odontogenic neoplasm characterized by simultaneous proliferation of both epithelial and mesenchymal component. It is most commonly seen between 1st and 2nd decades of life, the posterior aspect of mandible being the commonest site having slight male predilection. Herein, we report a case of ameloblastic fibroma in a comparatively higher age group of 35 year old female patient involving right mid face region.

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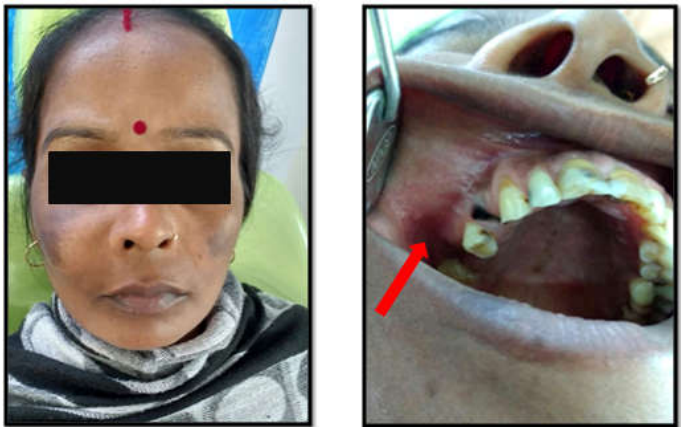
INTRODUCTION

Ameloblastic Fibroma (AF) is a very rare benign odontogenic neoplasm often designated as a mixed odontogenic tumour because of its characteristic feature of simultaneous neoplastic proliferation of both epithelial and mesenchymal component without formation of dentin or enamel matrix (Cohen *et al.,* 2004). They are likely to occur in young age group of patients, frequently diagnosed between 1st and 2nd decades of life (Cohen *et al.,* 2004; Ronald *et al.,* 1982) involving most commonly the posterior aspect of mandible, with slight male predilection (M:F=1.4:1) (Cohen *et al.,* 2004; Takeda, 1999). AFs usually exhibit relatively slower growth pattern than other odontogenic neoplasms. They do not generally tend to infiltrate between bony trabeculae, but instead, enlarges by gradual expansion, leading to a relatively smooth periphery of the lesion (Shafer *et al.*). Radiographically the tumour appears in the form of either unilocular or multilocular radiolucency with relatively smooth outline or often with sclerotic border. The lesion is most oflen associated with unerupted teeth. Microscopically, the lesion is composed of both ectodermal and mesenchymal component. The ectodermal component comprises of scattered islands of epithelial cells predominantly in long finger like strands or cords or often in rosette or nest like manner, characterized by peripheral cuboidal or columnar type of hyperchromatic epithelial cells resembling primitive odontogenic epithelium. Stellate reticulum like cells are very frequent but if found, they are found in larger nests. Mitosis is not a characteristic feature of this tumour. The mesenchymal component is formed of primitive connective tissue closely resembling those of dental papilla along with occasional paucity of blood vessels and juxtaepithelial hyalinization causing resemblance to dysplastic dentin. Microscopic differential diagnosis includes odontogenic myxoma and odontogenic fibroma (Mosqueda-Taylor *et al.,* 2011). Conventionally, AFs are treated by enucleation with curettage of surrounding normal bone while more aggressive lesions require a radical approach (Kulkarni *et al.,* 2013). Based on the clinicopathological, radiological, and histopathological features, a unique case of Ameloblastic fibroma involving the right maxilla of 35 years old female patient has been discussed herewith.

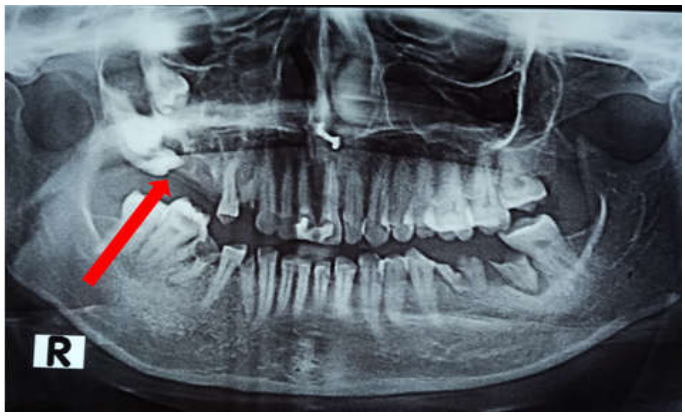
CASE REPORT

A 35 years old female patient reported to the Department of Oral and Maxillofacial Pathology with a chief complaint of a diffuse swelling in the right mid face region since one year. The swelling was progressive and gradually increasing in size with evidence of disturbance of vision in the right eye for last two months. Extraoral examination revealed a diffuse swelling extending onto zygomatic arch, mostly involving the right mid face region, with obliteration of the right nasolabial fold. Intraoral examination revealed presence of a fairly defined bony hard swelling in the right maxillary region extending from 14 to 18 region, with labial and buccal cortical plate expansion. And the overlying mucosa was apparently normal. OPG revealed presence of multilocular radiolucency in the right maxillary region with impacted tooth. Advanced radiographic technique like contrast computed tomography was done thereafter, to know the extent of the lesion and it showed hyperdense mass, measuring about 2.84cm×1.83 cm in

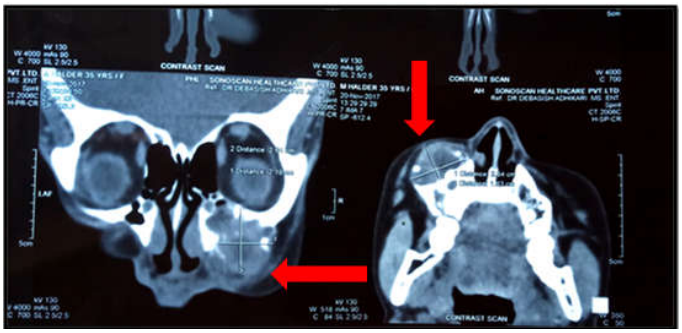


(a) (b)

Fig. 1(A). Extraoral photograph of patient showing diffuse swelling in the right mid face region ; (B) Intraoral photograph of patient showing a fairly defined bony hard swelling in the right maxillary region extending from 14 to 18 region

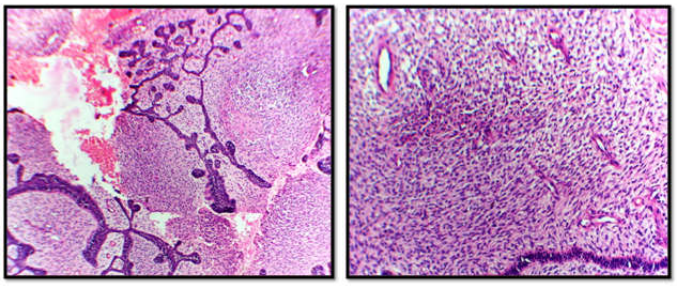


(a)



(b)

Fig. 2(A). OPG revealed the presence of multilocular radiolucency in the right maxillary region with impacted tooth ; (B): contrast computed tomography showing hyperdense mass in the right upper gingio-buccal sulcus with erosion of right maxillary alveolus and anterior wall of right maxillary sinus



(A) (B)

Fig 3. (A & B). Low power (H & E ,10X) shows odontogenic islands in cellular connective tissue and myxoid type of connective tissue stroma comprising of spindle shaped and angular cells

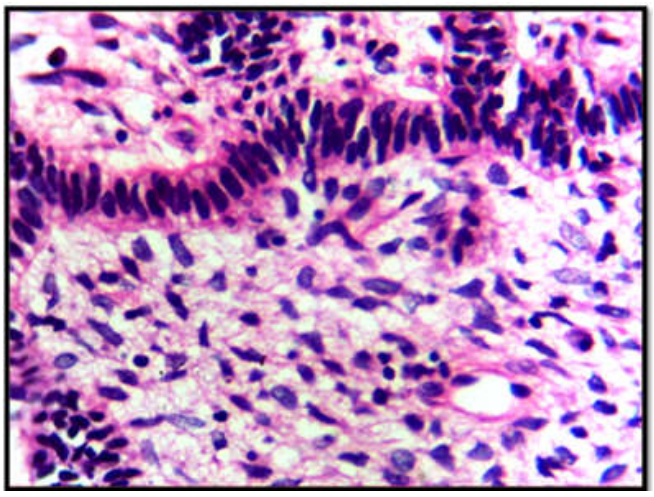


Fig. 4. High powe (H&E, 40X) shows tall columnar ameloblast like cells with reversal of polarity

the right upper gingivo-buccal sulcus with erosion of right maxillary alveolus and anterior wall of right maxillary sinus. Primarily, after clinical and radiological evaluation, the provisional diagnosis was in favour of odontogenic tumor.The lesion was enucleated with curettage of the surrounding normal bone. Microscopically, the lesion showed proliferation of islands of odontogenic epithelium consisting of tall columnar ameloblast like cells having hyperchromatic nuclei characterized by reverse polarity while mesenchymal components comprised of evenly distributed plump ovoid and stellate cells within a loose myxoid connective tissue. No atypia was observed. Hence the final diagnosis of Ameloblastic Fibroma was established.

DISCUSSION

Ameloblastic fibroma is a true mixed neoplasm comprising of both epithelial and mesenchymal components. It generally affects younger age group of patients, especially seen in the first two decades of life (Reichart *et al.,* 2004; Shafer *et al.,* 2006). The lesion most commonly occurs in the posterior part of mandible (Takeda *et al.,* 1999). Males are more prone to be affected than females, who are usually diagnosed at an early age (Cohen *et al.,* 2004). The case under discussion was of a 35 years old female patient which is contrary to the usual findings. The tumor generally is asymptomatic and is often diagnosed upon routine radiological examination (Cohen *et al.,* 2004; Tomich, 1999; Slootweg, 1981; Trodahl, 1972). But our case revealed a gradually increasing diffuse swelling over the right mid face region with disturbance in vision of the right eye since two months, along with cortical plate expansion. The lesion is often associated with impacted tooth, as in our case too, the radiographic image supported presence of impacted tooth (Trodahl, 1972 Regezi *et al.,* 1978; Philipsen *et al.,* 1997). Radiographically, AFs appear as unilocular or multilocular radiolucency with well defined borders (Chen *et al.,* 2007). These lesions may be confused with dentigerous cyst at the initial phase because they are often associated with impacted teeth (Cohen *et al.,* 2014; Tomich *et al.,* 1999; Hansen and Ficarra, 1988). Particularly, in this case we found multilocular radiolucency with impacted tooth. Based upon clinical and radiological finding, the case was provisionally diagnosed as Odontogenic Neoplasm or Cyst. And the differential diagnosis of the lesion includes ameloblastoma, odontogenic myxoma, keratocystic odontogenic tumor, central giant cell lesion,ameloblastic fibrosarcoma (Kobayashi *et al.,* 2005; Trodahl *et al.,* 1972; Philipsen *et al.,* 1997; Kim *et al.,*

2012; Vasconcelos *et al.,* 2009). Light microscopically the present case revealed connective tissue stroma that recapitulates dental papilla, composing of stellate shaped, spindle shaped and angular cells with little collagen, imparting a myxomatous appearance. The epithelial component comprises of odontogenic epithelium, mainly tall columnar ameloblast like cells, characterized by basophilic nuclei and little cytoplasm, arranged in cord or nest like fashion. This histopathological finding is totally supported by the authors of previous studies (Hansen and Ficarra, 1988). Thereafter the case was finally diagonosed as Ameloblastic Fibroma.Absence of mitotic figures helped us to differentiate it from Ameloblastc Fibrosarcoma. Thorough curettage of the lesion alongwith the affected teeth is the treatment of choice (Kulkarni *et al.,* 2013). Recurrence rate and malignant transformation of this lesion is recorded as relatively low in case of older patients(more than 22 years of age) where odontogenesis is completed (Philipsen *et al.,* 1997; Hansen *et al.,* 1988; Leider *et al.,* 1972; Altini *et al.,* 1985; Neville *et al.,* 2011).

Conclusion

AF is a rare tumor with good prognosis. However, patients should be followed up with great care after treatment and periodic special monitoring should be done in cases of recurring lesion which has higher propencity of malignant transformation. It will be definitely imperative to further refine our understanding of the lesion both clinically and histopathologically to render better treatment plan to patients.

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