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 often 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
tissue stroma comprising of islands in cellular connective tissue and myxoid type of connective tissue. 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 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 gingivo-buccal sulcus with erosion of right maxillary alveolus and anterior wall of right maxillary sinus.

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.

Fig 4. High power (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.,
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 propensity 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.

Acknowledgments: We would like to express our sincere thanks to Prof. (Dr.) R. R. Paul, Prof (Dr) S. Kundu, Dr Neha Shah and Dr Rudra Prasad Chatterjee for their immense help and relentless support.

Conflicts of interest: The authors of this manuscript declared that they have no conflicts of interest, real or perceived, financial or nonfinancial in this article.

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