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CASE STUDY

AMELOBLASTIC FIBROMA IN A 17- YEAR- OLD FEMALE: HAMARTOMA OR TRUE NEOPLASM? A CASE REPORT WITH REVIEW OF LITERATURE

***Neha Shah**

Department of Oral & Maxillofacial Pathology, Gurunanak Institute of Dental Sciences & Research, India

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ABSTRACT

Ameloblastic fibroma is an uncommon benign odontogenic tumour showing proliferation of both epithelial and mesenchymal components. It is usually seen in first two decades of life and posterior part of mandible is the commonest site involved. It is mostly associated with impacted or congenitally missing teeth. There is considerable debate as to whether it is a hamartoma or a true neoplasm. Herein, we report a case of ameloblastic fibroma in a 17- year old female patient involving the mandible and associated with impacted and congenitally missing teeth. The tumour showed aggressive features which were suggestive of a true neoplasm but age of the patient was corroborative to a hamartoma.

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INTRODUCTION

Ameloblastic fibroma (AF) is an uncommon, slow growing, benign, locally invasive, mixed odontogenictumor accounting for 1.5%-4.5% of all odontogenic neoplasms involving the jaw bones (Kulkarni *et al*., 2013; Jindal and Bhola, 2011; Gupta *et al*., 2011; Budharapu *et al*., 2014; Reichert and Philipsen, 2004; Cawson, Fifth edition). In 1891, Kruse first described AF and later, Thoma and Goldman in 1946 classified it as a separate entity (Kulkarni *et al*., 2013; Jindal and Bhola, 2011; Gupta *et al*., 2011; Reichert and Philipsen, 2004; Ealla *et al*., 2015). It is commonly seen in young adults, mostly in the first and second decade (Kulkarni *et al*., 2013; Jindal and Bhola, 2011; Budharapu *et al*., 2014; Reichert and Philipsen, 2004; Ealla *et al*., 2015; Neville *et al*., 2009). The youngest patient reported is a 7 week old infant (Gupta *et al*., 2011; Ealla *et al*., 2015; Gupta *et al*., 2010; Costa *et al*., 2011). It has slightly higher predilection for occurrence in males (M:F = 1.4:1) (Kulkarni *et al*., 2013; Jindal and Bhola, 2011; Reichert and Philipsen, 2004; Ealla *et al*., 2015). It is mainly seen as an intraosseous lesion, though few peripheral cases have also been reported (Kulkarni *et al*., 2013; Reichert and Philipsen, 2004). Mandible is affected more often than maxilla in 3.1:1 ratio (Jindal and Bhola, 2011; Reichert and Philipsen, 2004; Ealla *et al*., 2015). Clinically, small lesions are usually asymptomatic but large ones present as painless, slow growing swelling causing expansion of the jaws (Kulkarni *et al*., 2013; Jindal and Bhola, 2011; Gupta *et al*., 2011; Reichert and Philipsen, 2004; Cawson Fifth edition; Ealla *et al*., 2015; Neville *et al*., 2009; Costa *et al*., 2011). AF is mostly associated with impacted, uneruptedor congenitally missing teeth (Kulkarni *et al*., 2013; Jindal and Bhola, 2011; Gupta *et al*., 2011; Reichert and Philipsen, 2004; Ealla *et al*., 2015). Around 20% of cases are discovered accidentally duringroutine radiographic examination (Kulkarni *et al*., 2013; Jindal and Bhola, 2011; Reichert and Philipsen, 2004). Radiographically it appears as a well-defined unilocular (when small) or multilocular (when large) radiolucency with smooth sclerotic border (Kulkarni *et al*., 2013; Jindal and Bhola, 2011; Gupta *et al*., 2011; Reichert and Philipsen, 2004; Cawson, Fifth edition; Ealla *et al*., 2015; Neville *et al*., 2009; Gupta *et al*., 2010; Costa *et al*., 2011). AF must be differentiated clinically and radiologically from ameloblastoma, odontogenicmyxoma, dentigerous cyst, odontogenickeratocyst and central giant cell granuloma. Histologically - islands, strands, cords, nests or cauliflower like proliferations of odontogenicepithelium are seen in a loose but cellular, fibromyxoid connective tissue stroma containing plump fibroblasts and scanty collagen fibres resembling immature dental papilla. The strands show double or triple layer of cuboidal cells resembling dental lamina, whereas islands, cords or nests are surrounded peripherally by columnar ameloblast like cells enclosing stellate reticulum like cells in the centre (Kulkarni *et al*., 2013; Jindal and Bhola, 2011; Reichert and Philipsen, 2004; Cawson *et al*., Fifth edition;

Corresponding author:* **Neha Shah,
Department of Oral & Maxillofacial Pathology, Gurunanak Institute of Dental Sciences & Research, India.

Ealla *et al*., 2015; Neville *et al*., 2009). Histological variants of AF include i) granular cell type ii) papilliferous AF iii) ameloblastoma in association with AF iv) cystic AF v) ameloblasticfibrodentinoma and vi) ameloblastic fibro-odontoma (Kulkarni *et al*., 2013; Reichert and Philipsen, 2004; Ealla *et al*., 2015). Generally, enucleationof the lesionand removal of affected tooth is the treatment of choice for AF (Kulkarni *et al*., 2013; Gupta *et al*., 2011; Cawson *et al*., Fifth edition; Ealla *et al*., 2015; Gupta *et al*., 2010). High recurrence rate of 18% to 43.5% has been reported so long term follow up is necessary to look for recurrences and/or malignant transformation into ameloblasticfibrosarcoma (Kulkarni *et al*., 2013; Gupta *et al*., 2011; Budharapu *et al*., 2014; Reichert and Philipsen, 2004; Cawson *et al*., Fifth edition; Ealla *et al*., 2015; Neville *et al*., 2009).

Case report

A 17- year- old female patient reported to the Department of Oral & Maxillofacial Pathology of Dr. R. Ahmed Dental College & Hospital, Kolkata with a swelling in the lower jaw on the left side since twelve months. The swelling was small initially and it had grown gradually with time to attain the present dimensions. Extra-oral examination revealed a diffuse swelling in the left mandibular body region measuring about 2.0 cm x 1.5 cm with smooth, normal overlying skin (Figure 1). On palpation, it was hard, non-compressible, non-fluctuant and non-tender. The submandibular lymph nodes were not palpable. Intra-oral examination revealed a swelling in the left side of the mandible measuring about 2.5 cm x 1.5 cm extending from left mandibular first premolar to second molar (34 to 37) region. The overlying mucosa was erythematous and ulcerated in the buccal gingiva around left mandibular first and second premolar. Tooth indentation of the opposing left maxillary second premolar was present on the growth corresponding to edentulous space of 35 (Figure 2). On palpation it was firm in consistency, non-tender and non-fluctuant with expansion of both buccal and lingual cortical plates. Left mandibular second premolar was missing. Past dental history andmedical history were unremarkable. Orthopantomogram (OPG) showed a large, multilocular radiolucent lesion with sclerotic border involving the body, angle and ramus of mandible, extending anteriorly from 33 region and posteriorly upto 38 region. Left mandibular second premolar was missing whereas left mandibular third molar and deciduous left mandibular second molar were impacted. 75 was pushed towards the lower border of mandible with its crown towards the root of 36. There was slight root resorption near the apex of 34 and complete root resorption of 75 (Figure 3). Based on the clinical and radiological findings, a presumptive preoperative diagnosis of odontogenictumorwas made. Our differential diagnosis included dentigerous cyst and odontogenickeratocyst.

After informed consent from the patient, an incisional biopsy was performed under local anaesthesia from representative site.Section stained with hematoxylin and eosin, revealed the presence of scattered islands of proliferating odontogenic epithelial cells in a variety of patterns like rosettes, long finger like strands, nests and cords interspersed in a primitive connective tissue stroma which closely resembled the dental papilla consisting of few delicate collagen fibres with plump fibroblasts (Figure 4). Higher magnification (40x) revealed presence of tall columnar ameloblast like cells in the periphery of the islands along with loosely arranged cells resembling stellate reticulum in the centre (Figure 5). No hard tissue structures or mitotic figures were evident. The overall histological features were corroborative to the diagnosis of AF. Thereafter, thepatient was referred to the department of Oral & Maxillofacial Surgery for further surgical management where enucleation with curettage of surrounding bone and removal of 33 to 38 along with impacted 75 was done. The patient did not report back for further follow up.

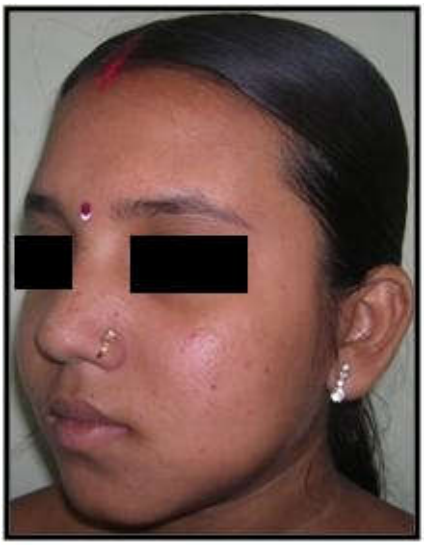

Figure 1. Extraoral photograph showing diffuse swelling in left mandibular body region

Figure 2. Intraoral photograph showing swelling in 34 to 37 region with erythema and ulceration of overlying mucosa in buccal gingiva of 34 & 35 along with indentation of opposing maxillary teeth

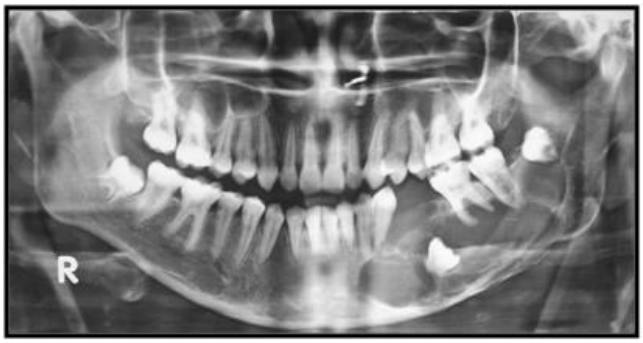

Figure 3. Orthopantomogram showing multilocular radiolucency with sclerotic border extending from 33 to 38 region along with missing 35, impacted 38 and 75 pushed towards lower border of mandible showing complete root resorption

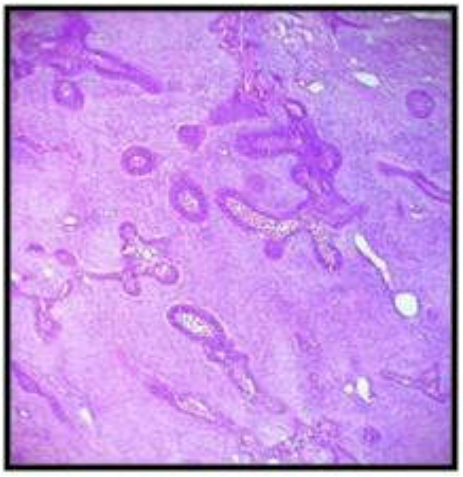

Figure 4. Low power photomicrograph (H&E, 10x) showing scattered islands, finger like strands and cords of odontogenic epithelial cells interspersed in primitive connective tissue stroma

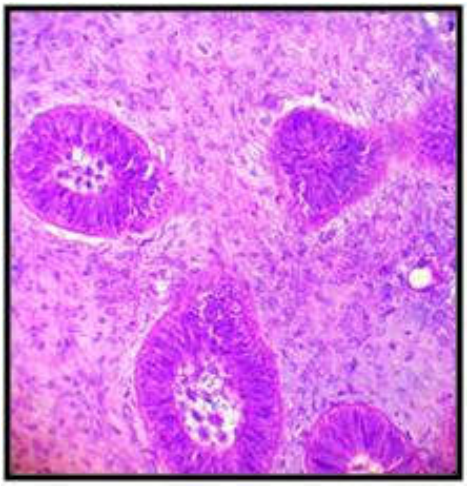

Figure 5. High power photomicrograph (H&E, 40x) showing presence of tall columnar ameloblast like cells in periphery of the island with loosely arranged stellate reticulum like cells in the centre. Primitive connective tissue stroma with few collagen fibres and plump fibroblasts can also be noted

DISCUSSION

WHO has definedameloblastic fibroma as a tumour composed of odontogenicectomesenchyme resembling the dental papilla with epithelial strands and nests resembling dental lamina and enamel organ, without hard tissue formation (Gupta *et al*., 2011). The nature and biological behaviour of AF is still not clearly understood. The 1992 WHO classification of odontogenic tumours did not include the definition ofameloblastic fibroma as a separate entity. It was listed with "related lesions," which alsoincluded ameloblasticfibrodentinoma and ameloblasticfibroodontoma. The suggested definition for this group of lesion was "neoplasm composed ofproliferating odontogenic epithelium embedded in a cellular ectomesenchymal tissue that resembles the dental papilla and with varying degree of inductive change and dental hard tissue formation". (Jindal and Bhola, 2011; Budharapu *et al*., 2014) Accordingly Cahn and Blum had postulated the "continuum concept" wherein it was assumed that an ameloblastic fibroma would, overtime, mature and finally result in the formation ofodontoma. (Jindal and Bhola, 2011; Gupta *et al*., 2011; Budharapu *et al*., 2014; Reichert and Philipsen, 2004; Ealla *et al*., 2015) This theory was refuted because 1) recurrent cases of AF never showed further steps of differentiation into dental hard tissue forming odontogenictumor of more advanced histodifferentiation. 2) AFs are known to occur at ages beyond completion of odontogenesis, that is, after the age of 20 years (Jindal and Bhola, 2011; Reichert and Philipsen, 2004). Hence, there is considerable debate and confusion as to whether AF represents an anomalous hamartomatous growth or is a true benign neoplasm. Recently it has been proposed that two variants of AF exist: neoplasm and hamartoma. These two are histopathologically undistinguishable. (Jindal and Bhola, 2011; Gupta *et al*., 2011; Budharapu *et al*., 2014; Reichert and Philipsen, 2004; Ealla *et al*., 2015) It is also suggested that AFs occurring after the age of20 years are true benign neoplasm whereas those developing during the period of odontogenesis may represent hamartomatouslesions. The hamartomatous variant has inductive potential whereas the neoplastic sub-type lacks inductive capabilities. Accordingly the neoplastic variant of ameloblastic fibroma and fibrodentinoma if left *in situ* does not differentiate further whereas the non-neoplastic, hamartomatous lesion is capable of developing into ameloblastic fibro-odontoma and differentiating further into complex odontoma. This latter line of development has been termed as complex odontoma line. (Jindal and Bhola, 2011; Reichert and Philipsen, 2004) In the present case, the tumour was seen in a female patient, contrary to the reported slightly higher prevalence for occurrence of AF in males. (Kulkarni *et al*., 2013; Jindal and Bhola, 2011; Reichert and Philipsen, 2004; Ealla *et al*., 2015) Our case was associated with congenitally missing left mandibular second premolar and impacted third molar concomitant with the findings of other authors. (Kulkarni *et al*., 2013; Jindal and Bhola, 2011; Gupta *et al*., 2011; Reichert and Philipsen, 2004; Ealla *et al*., 2015) Posterior part of mandible was involved in this case which is the most frequently affected site reported in literature. (Kulkarni *et al*., 2013; Jindal and Bhola, 2011; Gupta *et al*., 2011; Budharapu *et al*., 2014; Reichert and Philipsen, 2004; Cawson, Fifth edition; Ealla *et al*., 2015; Neville *et al*., 2009; Gupta *et al*., 2010) Radiologically, it presented with a multilocular radiolucency having sclerotic border extending from 33 to 38 region with impacted 38 and unerupted 75 showing complete root resorption. Most likely the growth must have pushed the 75 towards the inferior border of mandible and caused its root resorption. This case thus highlights the aggressive nature of the tumour. Further, our case is interesting as the age of the reported patient was 17 years but there was no evidence ofodontogenic differentiation in the tumour mass though it had been of long standing duration. Hence, it was difficult to determine whether it was a true neoplasm or a hamartoma. The age of the patient was corroborative to a hamatomatous growth but no evidence of histodifferentiation into dental hard tissues in the tumour was suggestive of neoplastic lesion. Enucleation with curettage of surrounding bone and removal of involved teeth was done as consensus existsregarding conservative management for AF in literature. (Kulkarni *et al*., 2013; Gupta *et al*., 2011; Cawson *et al*., Fifth edition; Ealla *et al*., 2015; Gupta *et al*., 2010)

Conclusion

It is essential to differentiate AF from other tumours in view of its neoplastic nature, possibility of recurrence and malignant transformation potential. So, despite consensus for conservative treatment, long term follow up is necessary.

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