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Case Report

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A RARE CASE OF AMELOBLASTIC FIBROMA IN THE MAXILLA: A CASE REPORT

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ABSTRACT

Ameloblastic fibroma (AF) is an extremely rare true mixed benign tumour that can occur either in the mandible or maxilla. It is believed to be a true mixed tumour, in which the epithelial and the ectomesenchymal elements are neoplastic. A 15-year-old girl had come to the department with a progressive swelling on the left side of her upper posterior jaw which occasionally bled. The swelling hindered occlusion and was fibrous in nature. Surgical excision of lesion was done under general anesthesia. Histopathologically, focal aggregates of lymphocytes were seen in stroma, suggestive of Ameloblastic Fibroma. Ameloblastic Fibroma enlarges by gradual expansion so that the periphery of the lesion often remains smooth. Conservative excision seems to be the treatment of choice. Large tumours may require a more aggressive approach, however. Ameloblastic fibroma is generally regarded as being less aggressive than the ameloblastoma, a feature which must be considered in the rational treatment and management of the patient with this tumour.

KEYWORDS: - Ameloblastic fibroma, Tumours of jaws, Conservative surgical excision, General anesthesia.

INTRODUCTION

Ameloblastic fibroma and related lesions are defined as "Neoplasms composed of proliferating odontogenic epithelium embedded in a cellular ectomesenchymal tissue that resembles the dental papilla, and with varying degrees of inductive change and dental hard tissue formation".[1] AF is defined by WHO as "consists of odontogenic ectomesenchyme resembling the dental papilla and epithelial strands and nests resembling dental lamina and enamel organ. No dental hard tissues are present."[2]

AF was first described by Krause in 1891. It usually occurs in the first two decades of life with a slight female predilection, causing delay in tooth eruption or altering the eruption sequence. Small tumours are asymptomatic, while larger ones produce significant swelling of the jaws.[3] Ameloblastic fibroma (AF) is an extremely rare true mixed benign tumour that can occur either in the mandible or maxilla.[4] It is frequently found in the posterior region of the mandible, often associated with an unerupted tooth.[5]

It is believed that ameloblastic fibroma may be a true mixed tumour, in which the epithelial and the ectomesenchymal elements are neoplastic. Lesions composed of similar elements, but in which inductive change has been resulted in the deposition of dentin alone or dentin plus enamel, are termed ameloblastic fibrodentinoma and ameloblastic fibro-odontoma, respectively.[6] This case is a rare presentation of ameloblastic fibroma in the maxillary posterior region.

CASE REPORT

A 15-year-old girl had come to the department of Pedodontics and Preventive Dentistry, New Horizon Dental College and Research Institute, Chhattisgarh, with a chief complaint of slowly progressive swelling on the left side of her upper posterior jaw which occasionally bled. The patient was apparently alright 2 years back after which she noticed a swelling on the left maxillary molar region. The swelling increased with time and increased to such an extent that there was difficulty in mastication. Her Medical history was unremarkable.

On extraoral examination there was a swelling on the left side of the face extending antero-posteriorly from the ala of the nose to the maxillary tuberosity and supero-inferiorly from the infra-orbital region to the corner of the lip (figure 1).

On intraoral examination, soft tissue swelling was seen antero-posteriorly on the left maxillary region extending from the over retained deciduous canine to the first permanent molar till the retromolar area measuring 3.5mm in length (figure 2). On palpation, the swelling was fibrous in nature and slightly tender. Both the teeth were Grade III mobile. Generalized gingival hyperplasia was seen in the mandible (figure 3).

Panoramic radiograph showed a unilocular radiolucent area with well-defined borders, involving the posterior aspect of the left maxilla. The permanent canine and both the premolars were submerged in the lesion (figure 4).

Fine needle aspiration did not yield any fluid ruling out a cystic lesion. Hence, surgical excision under general anaesthesia was performed.

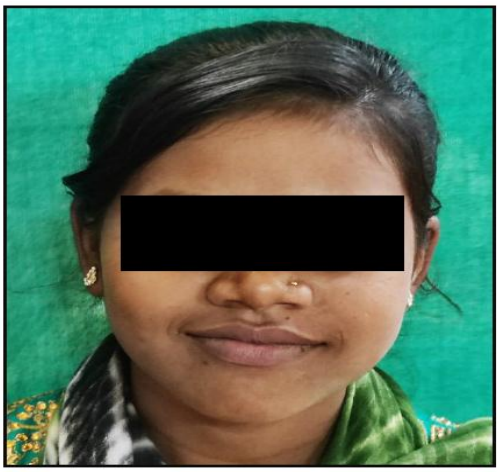

Fig. 1: Extraoral asymmetry of face seen on the left side due to the lesion.

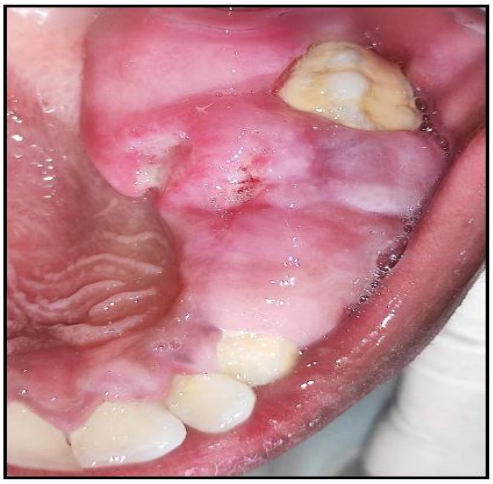

Fig. 2: Lesion seen intraorally in the maxilla extending from the deciduous canine to the retromolar region.

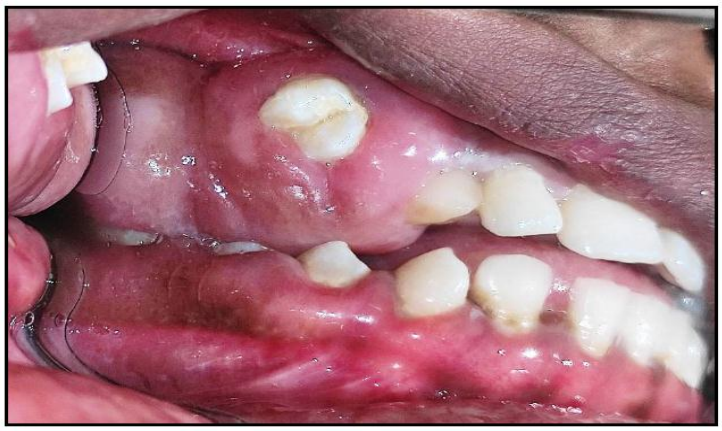

Fig. 3: Occlusal aspect of the lesion causing inability to Occlude and Difficulty in mastication.

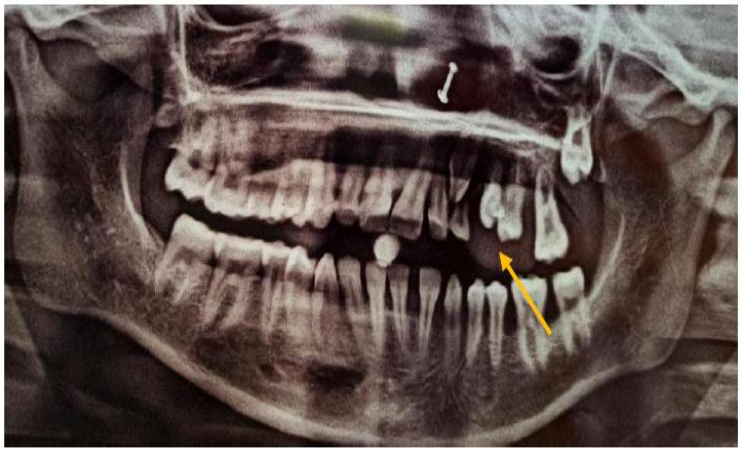

Fig. 4: OPG showing bone loss on the left Maxillary and Mandibular region. Arrowed area shows the lesion.

SURGICAL PROCEDURE

Under all aseptic precautions in the operation theatre, general anaesthesia was given to the patient. Local anaesthesia with adrenaline was given for tissue separation. Incision was made in the vestibular region and on the palatal side following the edges of the lesion. With the help of a periosteal elevator, the lesion was separated from the bone (figure 5). The teeth associated with the lesion were removed. Alveoloplasty was done to remove bony spicules. The teeth extracted were the permanent canine, the first and second premolars and the first molar. Electrocautery was used to remove any residual part of the lesion (figure 6). Haemostasis was achieved and suturing was done. The excised sample measuring 4.5cm x 3cm x 3cm was then sent for biopsy (figure 7).

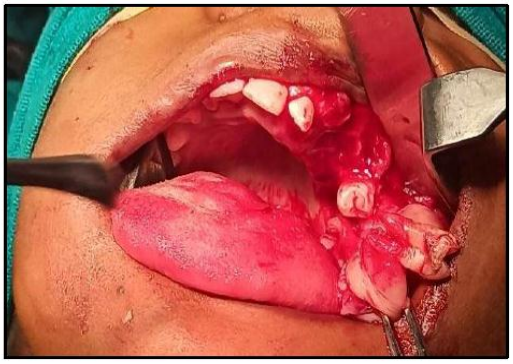

Fig. 5: Surgical removal of the lesion under general anaesthesia.

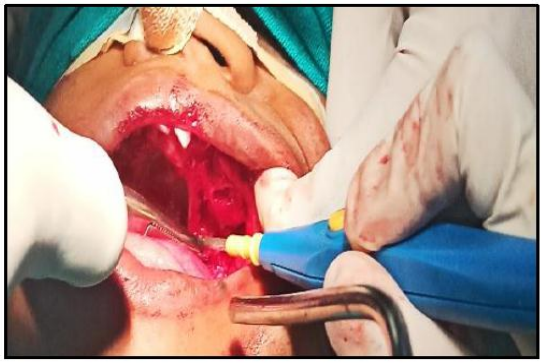

Fig. 6: Electrocautery used to remove any residual parts of the lesion which might cause recurrence.

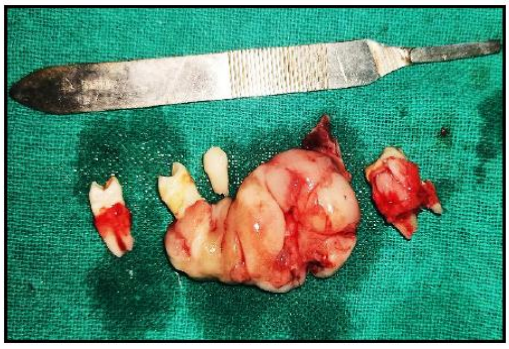

Fig. 7: Surgically excised mass.

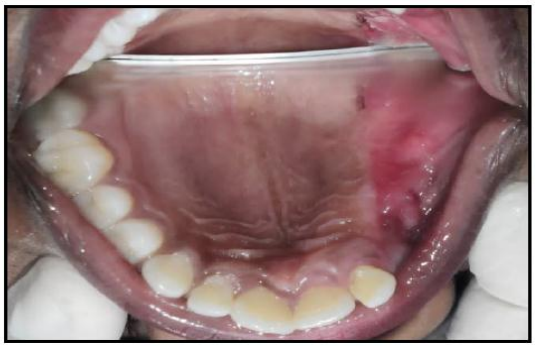

Fig. 8: 21 days post operative.

HISTOPATHOLOGY

Tissue sections showed portions of a spindle cell neoplasm composed of bland looking cells arranged in scattered pattern surrounded by abundant collagenised stroma with scattered islands and long cords and trabeculae of odontogenic epithelium lined by columnar cells, possessing vacuolated cytoplasm and mildly hyperchromatic nuclei with underlying areas composed of stellate reticulum like cells. Focal aggregates of lymphocytes were seen in stroma, suggestive of Ameloblastic Fibroma (figure 9, figure 10). There was no evidence of malignancy.

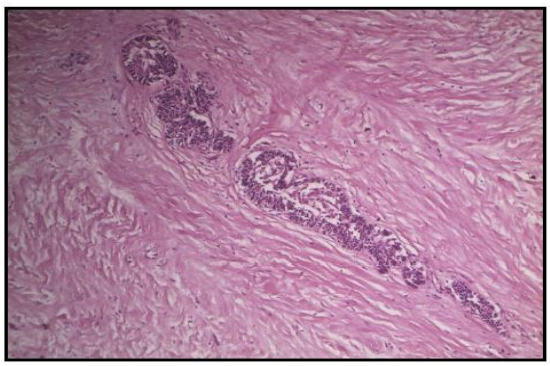

Fig. 9: Medium-power photomicrograph showing enamel organ differentiation of the epithelium and cellular fibrous connective tissue resembling dental papilla. H&E stain, original magnification X 100.

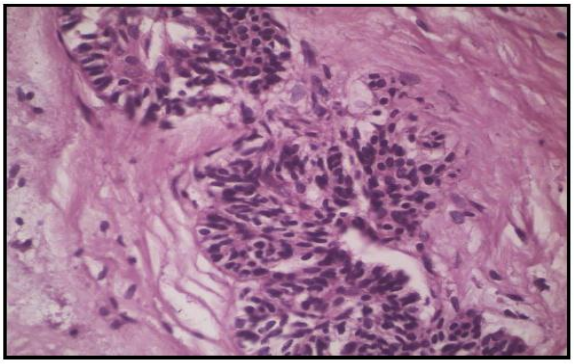

Fig. 10: High-power photomicrograph showing enamel organ differentiation of the epithelium and cellular fibrous connective tissue resembling dental papilla. H&E stain, original magnification X 200.

FOLLOW UP

On a follow up of 21 days (figure 8), 3 months and 6 months, the healing was uneventful. No residual tissues were seen.

DISCUSSION

Ameloblastic Fibroma is a relatively rare, benign neoplasm of the group of mixed odontogenic tumours. It represents only 2% of odontogenic tumors.[7] It is characterized by the simultaneous proliferation of epithelial and mesenchymal tissue, without the formation of dentin or enamel.[8] AF is a true mixed tumour in which both the epithelial and ectomesenchymal elements are neoplastic. The precise etiology of AF is not known. However, it is believed to arise de novo during a particular stage of odontogenesis, possibly as a result of overzealous elaboration of the basal lamina without further odontogenic differentiation.[9] Trodahl reported that in 58% of cases the presenting symptom is swelling.[10] Moreover, there is no significant gender difference in the frequency of AF or race predilection.[11] It has been reported as occurring at ages ranging from 6 months to 42 years, with an average from 14.6 to 15.5 years. The youngest patient was a 7-week-old infant reported recently.[12,13] Over 80% of these tumours occur in the mandible, usually in the canine-molar region.[14,15] Only 4 tumours have been reported in the anterior maxillary region.[16]

Ameloblastic fibroma exhibits somewhat slower clinical growth than the simple ameloblastoma and does not tend to infiltrate among trabeculae of bone.[17] Instead, it enlarges by gradual expansion so that the periphery of the lesion often remains smooth. It will frequently cause no complaint on the part of the patient and has been discovered accidentally during radiographic examination. Pain, tenderness or mild swelling of the jaw may induce the patient to seek aid from a dentist. Radiographically, no constant significant differences between the appearance of the simple ameloblastoma and that of ameloblastic fibroma are found.[17] The latter is manifested as a uni- or multi-locular, radiolucent lesion which has rather smooth outline, often with a sclerotic border, and which may or may not produce evident bulging of bone.[17]

Although Ameloblastic fibromas tend to separate readily from their bony walls and do not recur, a series from the Armed Forces Institute of Pathology reported a high recurrence rate of 43.5%.[17] Other researchers have reported a cumulative recurrence rate of 18% based on a review of the literature.[18] Conservative excision seems to be the treatment of choice. A modified block resection, rather than a curettage or simple excision, has been suggested. In their excellent review of mixed odontogenic tumours, Philipsen et al propose that the innocuous behaviour of the lesion does not justify aggressive initial treatment; meticulous surgical enucleation with close clinical follow-up should be sufficient.[18] Large tumours may require a more aggressive approach, however. Re-appearance of tumour may represent renewed growth of residual tumour rather than true recurrence. Such recurrent lesions should be treated more aggressively.[19]

CONCLUSION

Ameloblastic fibroma is generally regarded as being less aggressive than the ameloblastoma, a feature which must be considered in the rational treatment and management of the patient with this tumour. Conservative excision seems to be the treatment of choice.

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