Ameloblastic Fibroma of the Maxilla – A Rare Presentation

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ABSTRACT

Introduction: Ameloblastic fibroma is a rare tumour which occurs predominantly in children. It usually arises from the mandibular dentition although it can arise in maxilla. It is a benign tumor of mixed odontogenic origin. The common clinical manifestation is a slow-growing swelling and noneruption of teeth.

Case report: We present a case of ameloblastic fibroma of the maxilla in a 12 year old girl who presented to OPD with facial swelling. After detailed examination and investigations we diagnosed the case as a dentigerous cyst and the patient underwent enucleation through Caldwell Luc approach under general anaesthesia. Histopathological examination revealed ameloblastic fibroma. The patient is under long term follow-up with no signs of recurrence.

Conclusion: This case emphasizes the importance of careful differential diagnosis in ENT, while reporting a rare lesion and its atypical location.

Keywords: Odontogenic tumor, Ameloblastic fibroma, Maxilla, Dentigerous cyst

INTRODUCTION

Ameloblastic fibroma is a true mixed odontogenic tumour involving both epithelial and mesenchymal tissues. The tumor is of odontogenic origin and its occurrence is quiet rare accounting to only 2.5% of all tumors of this category.[1] It occurs predominantly in children and therefore remains an important diagnostic consideration.[2] It usually arises from the mandibular dentition although it can arise in maxilla.[1] It is characterized by simultaneous neoplastic proliferation of mesenchymal and epithelial components, with no formation of hard dental tissues.[3] Knowledge of the malignant potential of ameloblastic fibroma should assist in determining the management of these benign tumours, and may prevent malignant transformation to ameloblastic fibrosarcoma.[4]

Patients with ameloblastic fibroma of the maxilla usually present with intraoral findings including a mass on the anterior upper jaw, swelling of the alveolar process, and non-eruption of teeth. Microscopically, the lesion shows cords and islands of neoplastic odontogenic epithelium interspersed by neoplastic mesenchymal tissue that resembles the dental papilla. If lesions composed of similar elements are found with deposition of dentin then they are termed as ameloblastic fibrodentinoma and if dentin and enamel both are found then it is called ameloblastic fibroodontoma.[5]

Radiographically, ameloblastic fibroma appears as a well-defined, unilocular or multilocular radiolucent lesion, with sclerotic radiopaque margins. Ameloblastic fibroma may be initially identified in routine radiographic examination.[6]

The most appropriate treatment method for ameloblastic fibroma is still uncertain. A conservative approach is suggested by several authors.[7-10] However, tumors may recur following surgical removal and progress to malignancy.[4,6,7] Thus, long-term follow-up of ameloblastic fibroma is recommended.[4] This report describes an interesting case of ameloblastic fibroma that affected the maxilla of a young girl with cheek swelling.

CASE REPORT

A 12-year-old female patient reported to the Department of ENT with the complaint of a painless swelling of right cheek since 20 days associated with epiphora of the right eye since 20 days. External examination showed a diffuse swelling of the right maxilla. Intra-orally, a firm, non-tender swelling was noted in the buccal vestibule extending from the region of the right maxillary central incisor to the first premolar. Swelling also extended over the corresponding palatal surface. The right canine tooth was missing (Figure-2). The remaining ENT and general examination was normal.

CT scan was in favour of mixed dense soft tissue lesion showing cystic and solid components seen in the right maxillary sinus. Mild enhancement of solid components. Expansion of the sinus seen with thinning of walls. Tooth like structure seen in the superior part of the lesion. No obvious intra-orbital extension was seen. Possibilities included: Dentigerous cyst, Fungal granulomatous lesion and Retention cyst (Figure-1).

With these data a provisional diagnosis of dentigerous cyst was made. After surgical profile and pre anaesthetic check-up, tumor enucleation along with impacted tooth through Caldwell Luc approach under general anaesthesia was planned. After mucosal incision antero-inferior wall of maxillary sinus was found eroded by the lesion (Figure-2). After enucleation and proper hemostasis, the wound was closed by 3/0 catgut. The specimen was subjected to histopathological examination (Figure-3).

On histopathological examination, the specimen revealed islands of epithelial cells in ectomesenchyme resembling the dental papilla. The islands consist of a peripheral columnar ameloblastic cells with reverse polarity in fibromyxoid stoma. Some of the areas show a zone of hyalinization and formation of dentinoid at the periphery (Figure-4). Based on the histopathological examination, a final diagnosis ameloblastic fibroma was arrived at. Post operative healing was good and no sign of recurrence of AF was recorded during the follow-up period.

DISCUSSION

Mixed odontogenic tumors comprise a rare group of lesions with diverse histopathological types and clinical behaviour.

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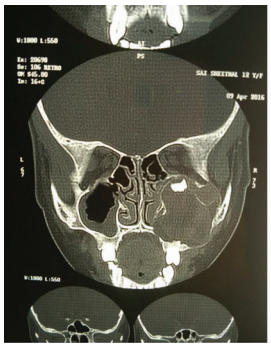
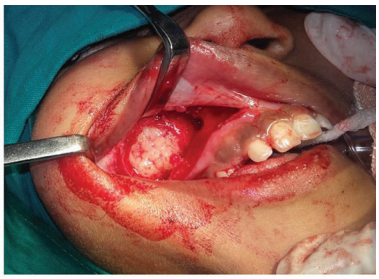

Figure-1: Showing CT image; **Figure-2:** Showing missing canine tooth and Caldwell Luc approach

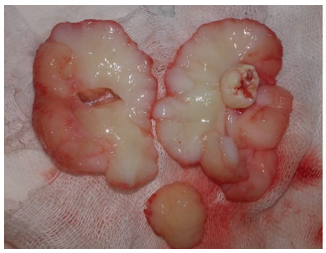
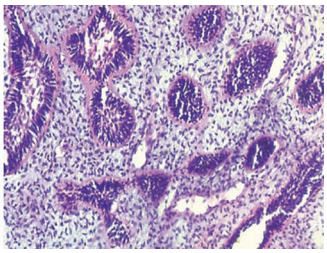

Figure-3: Cut section of the specimen showing the tooth; **Figure-4:** Histopathological slide

There is still confusion regarding the nature and relationship of these tumors and other related lesions in the existing literature. Mixed odontogenic tumors generally comprise of Ameloblastic fibroma (AF), Ameloblastic fibrodentinoma and ameloblastic fibro-odontoma.

AF has both epithelial and mesenchymal neoplastic proliferation.[8] This tumor is usually diagnosed between the first and second decades of life and the majority of cases of occur in the mandible.[6,9] A few cases of the AF have been reported in the maxilla.[9] The clinical manifestations of AF are not specific and the lesion is frequently discovered as an incident finding in a routine radiographic examination, such as cysts and other odontogenic tumors.[8] Radiographically, ameloblastic fibroma must be differentiated from other similar lesions such as ameloblastoma, dentigerous cysts, odontogenic keratocysts and fibrosarcoma ameloblastic.[4,10]

While uncommon, the possibility of malignant transformation of AF into ameloblastic fibrosarcoma has been reported by Chen etal.[6] and Kobayashi et al.[4] In those studies malignant change only occurred in the second recurrence. But according to Kousar et al.[7] rapid sarcomatous transformation of an AF occurred within 6 months. All authors agree that a long term follow-up is necessary.

Ameloblastic fibroma should generally be treated with conservative surgery. Only larger lesions or recurrent lesions should be treated with radical surgery. Recurrence rate is estimated to be around 33.3%, but there is a lower incidence among younger patients.[6] Therefore enucleation via Caldwell Luc approach was considered sufficient in this patient, due to her young age.

CONCLUSION

Ameloblastic fibroma is a rare tumor and has a good prognosis. AF and other related lesions should be kept in mind while diagnosing the swellings of the maxillofacial region in children and adolescence. After proper clinical, radiological and histopathological examination, conservative surgery is enough in most of the cases of AF. Patients must be kept under long term follow-up and special care should be taken for recurring tumors. The present case emphasizes the importance of careful differential diagnosis in ENT, while reporting a rare lesion and its atypical location.

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