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Ameloblastic fibro-odontoma

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ABSTRACT

INTRODUCTION: Ameloblastic fibro-odontoma (AFO) is a quite rare, mixed odontogenic tumour generally seen in the early stages of life. Frequent signs of this tumour are asymptomatic swelling, delayed tooth eruption and mixed radiological appearance within well-defined borders. Management of the lesion includes enucleation of the tumour and long-term follow-up.
PRESENTATION OF CASE: A 10-year-old girl was referred to our oral and maxillofacial surgery clinic with an incidental radiological finding of radiopaque mass in the posterior region of maxilla. OPG showed unerupted tooth bud of upper right second molar and was being prevented from eruption by the odontome. Under general anaesthesia, the lesion was enucleated and the permanent right upper second molar tooth bud removed.
DISCUSSION: Mixed odontogenic tumours are a group of rare and interesting lesions which can mislead the clinician to variety of differential diagnosis. Adequate clinical and radiological investigations, proper surgical excison, accurate histopathological diagnosis, and long term follow up will ensure the right treatment plan for the patient.
CONCLUSION: The possibility of a mixed rare tumour should be kept in mind by the clinician where they deal with the swellings of posterior maxilla in children. Histological assessment revealed a final diagnosis of ameloblastic fibro-odontoma.

1. Introduction

The ameloblastic fibro-odontoma (AFO) is a rare benign odontogenic lesion defined as a tumour with the general features of ameloblastic fibroma but that also contains enamel and dentine. According to the recent World Health Organization classification of Odontogenic Tumours published in 2005,[1] AFO belongs to the group of lesions with odontogenic epithelium with odontogenic ectomesenchyme, with or without hard tissue formation. Ameloblastic fibro-odontoma is defined by the World Health Organization as a neoplasm consisting of odontogenic ectomesenchyme resembling the dental papilla, epithelial strands and nest resembling dental lamina and enamel organ conjunction with the presence of dentine and enamel.

AFO is normally found in young patients, with no significant gender predilection. The two main complaints associated with AFO are swelling and failure of tooth eruption. Clinically, it presents as a painless swelling of the affected area, usually the posterior portion of the maxilla or mandible. Radiographs show a well-defined radiolucent area containing various amounts of radiopaque material of irregular size and form.

2. Case report

A 10-year-old girl was referred to oral and maxillofacial surgery department with an incidental radiological finding of radiopaque mass in the posterior region of maxilla. On inspection a mild swelling was present in the right posterior buccal region. On palpation lesion was firm in consistency, and gave the feeling of irregular surface. Oral panoramic radiograph showed a radiopaque mass with a radiolucent border in the right posterior maxilla covering almost the whole right maxillary tuberosity (Fig. 1). Considering the clinical and radiological picture the possible differential diagnosis was calcifying epithelial odontogenic tumour (CEOT), adenomatoid odontogenic tumour (AOT) and ameloblastic fibro-odontoma.

Under general anaesthesia with endotracheal intubation, a crevicular incision was made from the premolar region till the end of the maxillary tuberosity with a releasing incision vertically from the distal end of second premolar. A trapezoidal flap was elevated and a thin bone covering the surface of the lesion was removed (Fig. 2). The lesion was enucleated out with relative ease. Tooth bud attached to the follicle around the lesion was removed. The cavity

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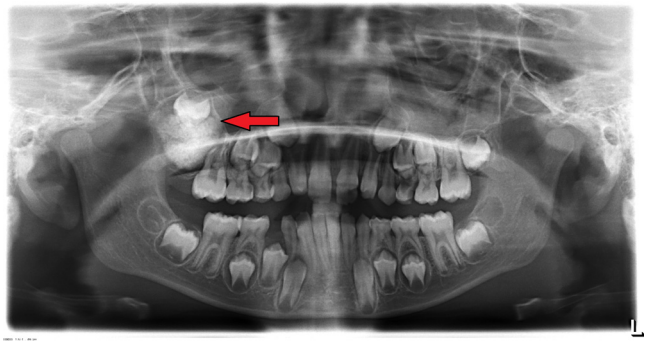

Fig. 1. Preoperative OPG.

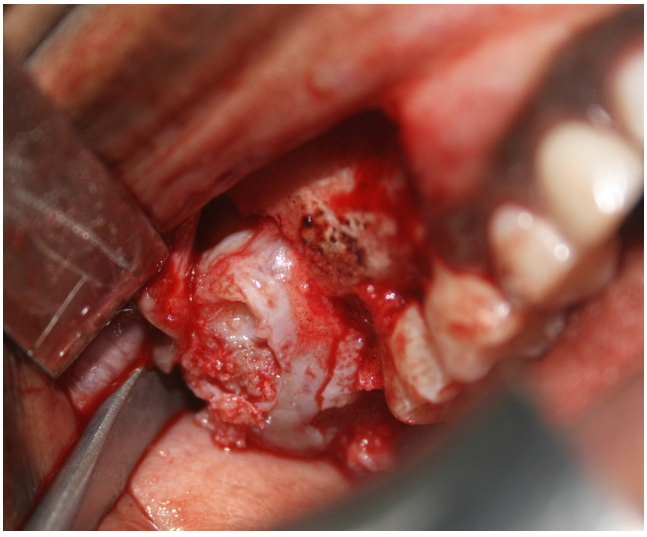

Fig. 2. Lesion exposed.

was irrigated carefully and the debris was removed. Fig. 3 shows the excised mass. The flap was repositioned in the same position and suturing was done with vicryl 3.0 suture material. Fig. 4 shows the postoperative OPG after 6 months.

3. Histopathology

On gross examination the specimen consisted of a hard tissue mass with a soft tissue attachment. The histopathologic examination of the soft tissue revealed numerous cords and follicles of odontogenic epithelium exhibiting peripheral ameloblast-like cells within primitive ectomesenchymal tissue (Fig. 5). Hard tissue examination exhibited conglomerate mass of enamel and dentine arranged in a disorganized pattern and in close relationship to the ameloblastic epithelium (Fig. 6).

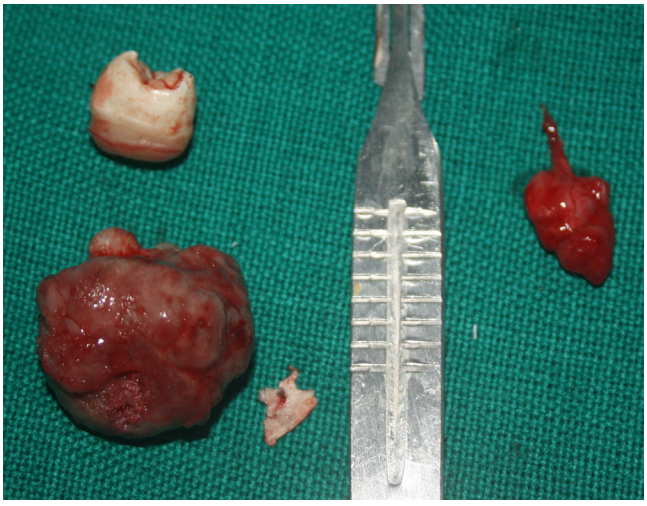

Fig. 3. Excised lesion.

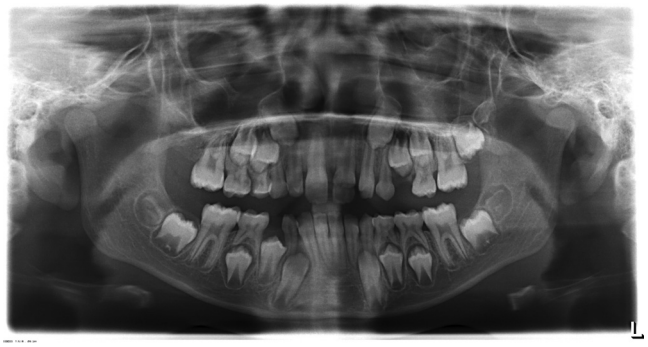

Fig. 4. Postoperative OPG after 6 months.

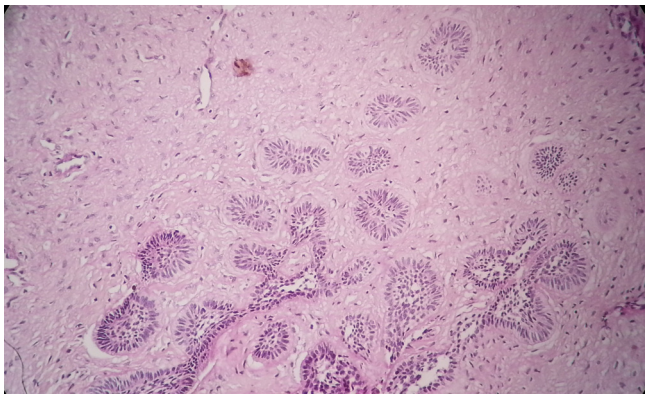

Fig. 5. Histopathology (40×) showing follicles of amelobalstic epithelium within a primitive ectomesenchyme.

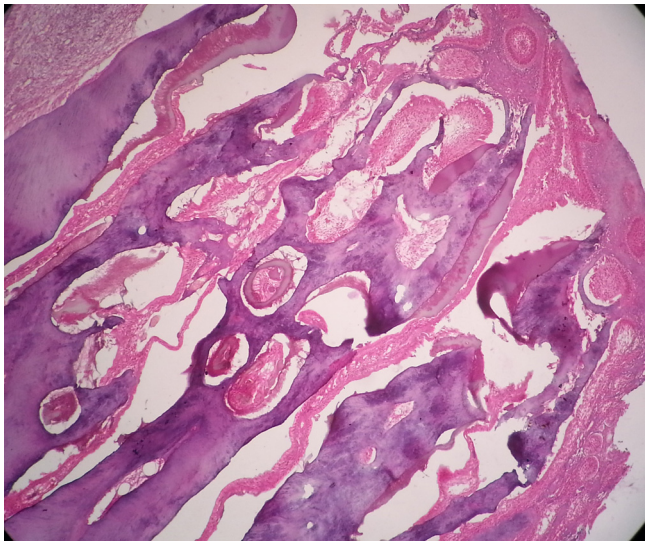

Fig. 6. Histopathology (10×) decalcified section showing areas of enamel and dentine in close relationship with the ameloblastic epithelium.

4. Discussion

Ameloblastic fibro-odontoma (AFO) is an uncommon mixed odontogenic tumour.[2] In a recent comprehensive study, AFO has been reported with a prevalence range of 0–3.4% within odontogenic tumours among different regions. Generally it is seen in the first and second decades of life, which might also be a characteristic of the lesion.[3,4] However, AFO may also occur at advanced ages.[5,6] Previously, Philipsen et al.[7] declared that the mean age of AFO cases falls when compared with ameloblastic fibroma (AF) and ameloblastic fibro-dentinoma, supporting the suggestion that age is a critical feature in AFO diagnosis.

Common signs and symptoms of AFO are asymptomatic swelling, delayed tooth eruption in the affected region and a well-defined mixed radiological appearance that is similar to other odontogenic neoplastic formations, such as immature complex odontoma, calcifying epithelial odontogenic tumour and adenomatoid odontogenic tumour. Nevertheless, final diagnosis is made according to microscopic evaluation demonstrating islands of odontogenic epithelium embedded in cell-rich ectomesenchyme similar to dental papilla. AFO can be differentiated from AF by the radiological appearance as well as through histological evaluation. It is also distinguishable from ameloblastic fibro-dentinoma not only because it shows dental structures resembling dentine, but also because it contains enamel-like tissues. Thus, the formation of AFO might be based on enamel matrix production, which is one of the most important features of the lesion.

The WHO (World Health Organization) classification describes AFO as a lesion similar to AF, also showing inductive changes that lead to the formation of both dentine and enamel. In addition, AF and AFO have been defined as hamartomatous lesions and are believed to be stages of odontoma formation.[8,9] This means that the aforementioned lesions should not be considered as distinct entities.

The treatment of AFO is associated with conservative surgical approach. Sporadic recurrences of AFO have been attributed to the inadequate surgical removal at the time of initial treatment. Some other reports demonstrated that a conservative enucleation is enough. There is a controversy in the literature regarding extraction or retaining the associated tooth bud in the case of AFO. Majority of the articles state that the associated tooth bud has to be removed in order to avoid recurrence.

5. Conclusion

Clinicians when dealing with radiopaque mass in posterior maxilla of children should keep the possibility of rare mixed odontogenic tumour in mind. Most of these benign lesions on complete excision heal well, but they need long-term follow-up.

Conflict of interest

None declared.

Ethical approval

This case report is the surgical treatment of an odontogenic tumour in the maxilla. The treatment of Amelobalstic fibro odontoma is surgical excision. The treatment plan was approved in the joint discussion by the maxillofacail surgeons, pedodontist and oral pathologist who are co authors for this paper.

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None.

Author contributions

Dr. Surej Kumar, Dr. Suvy Manuel and Dr. Sherin A. Khalam were the surgeons, Dr. Sivakumar was the Oral Pathologist and Dr. Jyothi Issac was the Pedodontist for the study. In addition, Dr. Surej Kumar was also involved in data collection, Dr. Suvy Manuel in data analysis and Dr. Sherin A. Khalam for review of literature. Dr. Kannan Venugopal was responsible for writing the article. Dr. Suvy Manuel and Dr. Kannan Venugopal were responsible for the manuscript preparation.

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