case report

Bilateral Ameloblastic Fibroma in A 6 Year Old Child: A Unique Case

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ABSTRACT

Ameloblastic fibroma (AF) is an extremely rare true mixed benign tumour that can occur either in mandible or maxilla, but is most frequently found in the posterior region of mandible. It is usually seen in the first two decades of life and is associated with tooth enclosure, hence causing delay in eruption or altering the dental eruption sequence. AF is found during routine radiographic evaluation but is clinically and radiographically similar to Ameloblastic fibro-odontoma and odontoma, which makes an accurate diagnosis mandatory as it may change the course of treatment. There has been lot of debate regarding the treatment for AF. We describe a case of **bilateral** *ameloblastic fibroma in the posterior region of mandible in 6 year old child which was treated by conservative curettage with admirable results.*

KEYWORDS

Ameloblastic fibroma, Curettage

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INTRODUCTION

Ameloblastic fibroma is defined as "Neoplasms composed of proliferating odontogenic epithelium embedded in a cellular ectomesenchymal tissue that resembles the dental papilla, with varying degree of inductive change and dental hard tissue formation(1). This tumour is reported to occur at an age ranging 6 months to 45(2) years, with an average of 14.6 to 15.5 years(3). The youngest patient reported is a 7-week old infant(4). It can appear in either the mandible or maxilla but Over 80% of tumours occur in the mandible(5). Only four cases of tumors in the maxillary anterior region have been reported.

This article presents an interesting case of a AF that had affected the mandible of a young child and was associated with unerupted mandibular teeth. The most remarkable finding however was its bilateral occurrence in mandibular molar area which is very rare.

CASE REPORT

A 6 year old child was referred to the department of Oral and Maxillofacial Surgery for evaluation of a painless bony hard swelling present bilaterally at mandibular body ramus area, which has been progressively enlarging for the past 6 months. Physical examination showed otherwise healthy child.

The intra oral examination revealed a large painless expansion of mandibular buccal and lingual cortical plates extending from the 1st permanent molar to the ramus of the mandible bilaterally. The posterior extension could not be palpated intra orally. All deciduous teeth and 1st permanent molar are in normal occlusion. The swelling was more prominent on buccal side with clinically healthy overlying mucosa. The bony cortex over the anterior border of the ramus was generally thin and found to be penetrated in a small area on anterior border of ramus. The rest of the head and neck examination was normal and medical history was unremarkable.

Panoramic radiograph shows bilateral radiolucencies. On right side, unilocular radiolucency extending from distal to tooth bud of permanent 2nd molar, to 1 cm below the coronoid notch. On left side multilocular radiolucency extending from distal to 1st molar to the coronoid notch pushing the inferior alveolar canal to the posterior border of ramus. Tooth bud of 2nd molar is missing on left side (Figure 1). Post Operative 6 month follow up radiograph shows excellent bone healing (Figure 2).

Histopathology shows parakeratinized stratified squamous epithelium overlying a densely collagenous connective tissue with rich cellularity. An ameloblastomatoid island with peripheral tall columnar cells exhibiting reversal of polarisation and hyperchromatic nuclei, and loosely arranged central polyhedral to stellate – shaped cells resembling stellate reticulum can be seen. Few strands and nest of resting odontogenic epithelium showing hyperchromatic nuclei and mild chronic inflammatory cell infiltrate (predominantly lymphocytes and plasma cells) is present (Figure 3, 4).

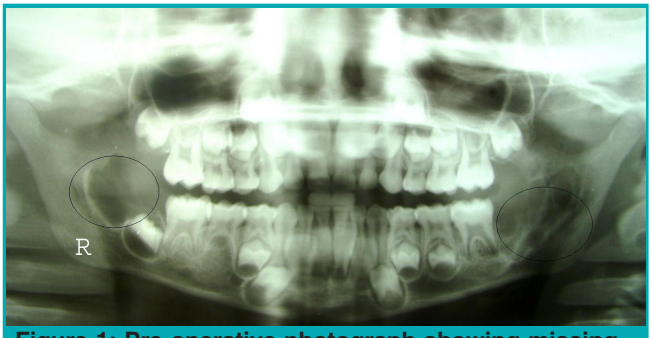

Figure 1: Pre-operative photograph showing missing tooth bud of left second molar along with bilateral radiolucencies

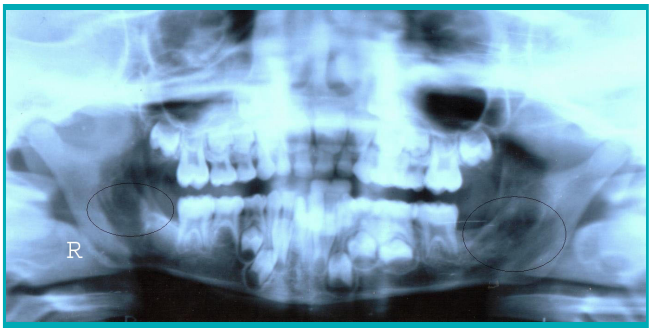

Figure 2. Post operative 6 month follow up radiograph shows excellent bone healing

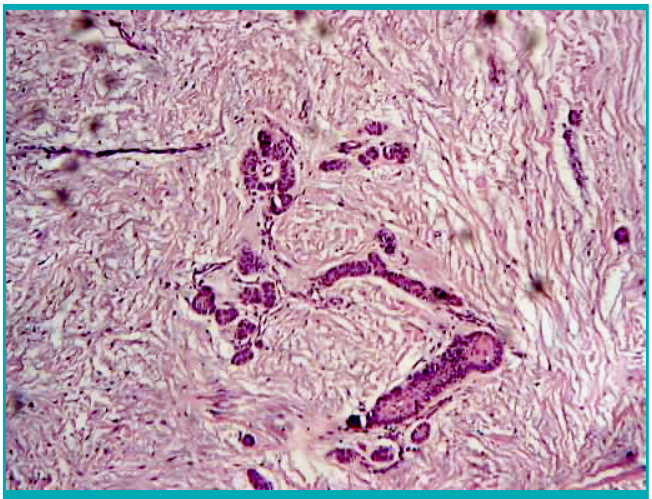

Figure 3: Ameloblastic fibroma containing islands of odontogenic epithelium (h & e stained original magnification x 10)

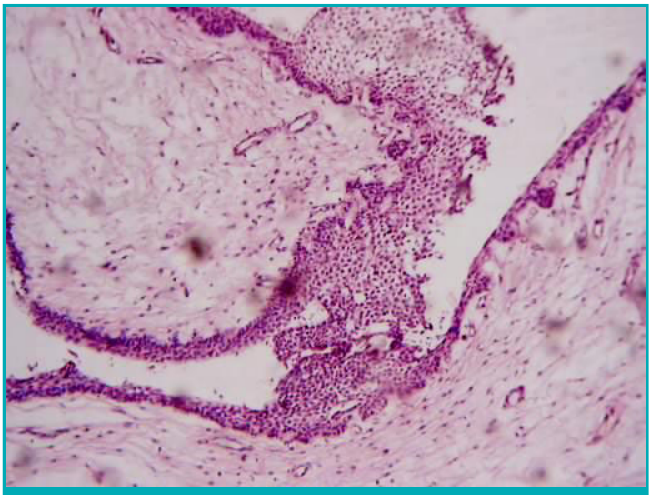

Figure 4: Ameloblastic fibroma containing odontogenic epithelium distributed in a cell rich primitive connective tissue stroma resembling dental papilla (h & e stained original magnification x 40)

DISCUSSION

Ameloblastic fibroma is a mixed intra osseous tumour of the odontogenic origin; a true biphasic tumour consisting of proliferating odontogenic epithelium in ectomesenchymal tissue that resembles the dental papilla. Both epithelial and mesenchymal tissues are neoplastic(6, 7).

AF is usually associated with enclosed teeth in the posterior region of the mandibular angle and /or ramus. The radiographic appearance may vary from a small unilocular lesion to an extensive multilocular lesion. Bone expansion and tooth dislocation are common findings. In the case reported, lesion is unilocular on left side and multilocular on right side and there is no tooth bud of 2nd permanent molar on left side. These findings make this lesion unusual.

Histologically, AF is true mixed tumour, as it has both mesenchymal and epithelial components with no associated calcified tissue. Ameloblastic fibrodentinoma is defined as "a neoplasm similar to Ameloblastic fibroma, but also showing inductive changes that lead to the formation of dentine."Ameloblastic fibro-odontoma is defined as "A lesion similar to Ameloblastic fibroma, but also showing inductive changes that lead to formation of both dentin and enamel." Dentinoma is defined as a very rare neoplasm composed of odontogenic epithelium and immature connective tissue and characterised by the formation of dysplastic dentin. It is thought that Ameloblastic fibrodentinoma, Ameloblastic fibro-odontoma and dentinoma are histologic variant in which there is formation of dentin or dentinoid tissue(8).

In WHO classification of tumours, the term Ameloblastic fibrodentinoma and dentinoma is used synonymously.

The treatment performed in the present case is simple surgical curettage and enucleation along with removal of associated tooth bud. Few authors prefer to do mandibular resection followed by immediate reconstruction. In this young 6 year old patient, resection will result in mutilation. So we preferred enucleation and curettage. However, in extensive recurrent lesion, block resection can be recommended followed by primary reconstruction.

Whatever the treatment may be, patients with AF must be followed up for long period to detect the early recurrence or development of Ameloblastic fibrosarcoma, which is the malignant counterpart of AF.

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