CASE REPORT

Ameloblastic fibroma in one-year-old girl

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ABSTRACT
Ameloblastic fibroma (AF) is a relatively rare, slow growing benign mixed odontogenic tumor, comprising of 1.5-4.5% of all odontogenic tumors. It is usually asymptomatic except for the eventual expansion of the jaw. AFs are most common in adolescents and young adults, mostly affecting the mandible as a well-defined uni or multilocular radiolucency. The effective surgical treatment includes enucleation and curettage of the surrounding bone and removal of the affected teeth. Although recurrence of AF is rare, a long term follow up is recommended. This report describes a 1-year-old girl with AF in the mandible and discusses its clinical, radiographic and histological findings.
Key words: **Ameloblastic fibroma, mandible, odontogenic tumour**

INTRODUCTION

Ameloblastic fibroma (AF) is a relatively uncommon, slow growing benign neoplasm of odontogenic origin which is characterized by simultaneous proliferation of both epithelial and mesenchymal tissues without formation of enamel or dentin.[1] It represents only 1.5-4.5% of all odontogenic tumors.[2] It was first described by Kruse in 1891 as cited by Edward Mosby.[3]

The precise etiology of AF is not known; however, it is believed to arise *de novo* during a stage of odontogenesis, possibly as a result of overzealous elaboration of the basal lamina without further odontogenic differentiation.[1] It consists of the group of odontogenic epithelium in dental papilla like background without dental hard tissue formation.[4,5]

CASE REPORT

A 1-year-old girl child was brought by her parents to our hospital with a complaint of swelling in the mandibular right posterior region. The mother of the child noticed the swelling since 2-3 months, which gradually increased in size. Her medical, surgical, family and social history were unremarkable. A systemic review was within normal limits and no medication had been taken for this swelling.

Access this article online	
Quick Response Code:	Website: www.jomfp.in
	DOI: 10.4103/0973-029X.110734

Intraoral examination showed a hard swelling in the right mandibular body, which was present in the canine to molar region. The overlying mucosa was intact and of normal colour. The buccal cortical plate expansion in the region of the swelling was detected [Figure 1].

There was no evidence of pus or blood discharge from the swelling. The radiographic examination (occlusal and lateral oblique view) showed a large well defined unilocular radiolucent lesion extending from right deciduous canine to second molar region. The borders of the lesion were well defined and corticated with thinning and expansion of the inferior border of mandible and buccal cortex. The lesion was associated with a displaced developing mandibular first molar towards inferior border of mandible while second molar was displaced posteriorly [Figures 2 and 3]. The clinical diagnosis of dentigerous cyst was made. Enucleation of the lesion was advised, but the parents refused the surgical treatment.

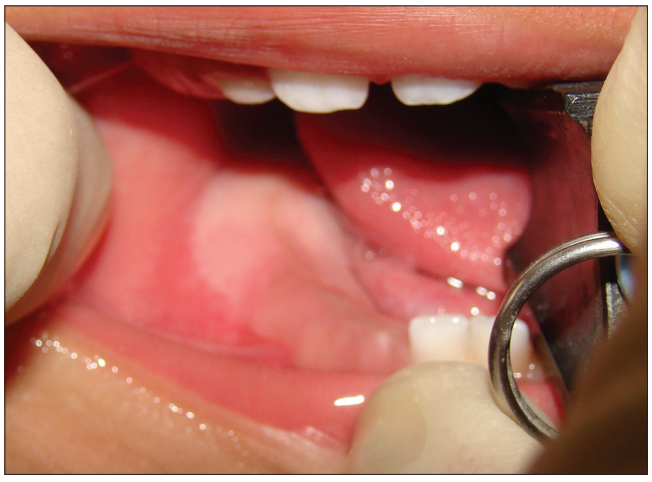

Figure 1: Intraoral photograph of the lesion

Again after 3 months the patient reported with a rapid increase in the size of the swelling. A proliferative growth was evident in the mandibular right posterior region extending from canine to molar region. Approximate size of the lesion was 3 cm × 2 cm which was soft and non-tender. Right submandibular lymphadenopathy was present. Taking into consideration the sudden rapid growth, which was proliferative a clinical suspicion of a malignant lesion, ameloblastic fibrosarcoma was considered, and an incisional biopsy was performed.

H and E stained sections showed a tumor mass made up of epithelial and mesenchymal components of odontogenic origin. The epithelial component consisted of multiple, sharply defined strands and islands which were bordered at the periphery by a layer of tall columnar cells resembling ameloblasts. The mesenchymal component is made up of primitive connective tissue consisting of closely interwining fibrils interspersed by large connective tissue cells closely resembling those of dental papilla. The histopathological diagnosis was AF [Figures 4 and 5].

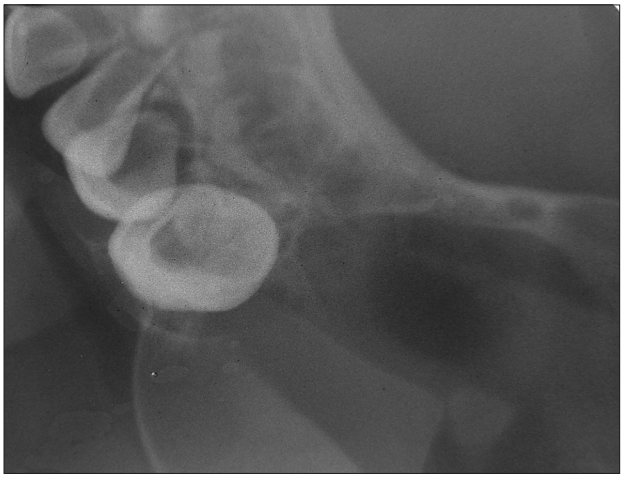

Figure 2: Occlusal view showing expansion of cortical plates

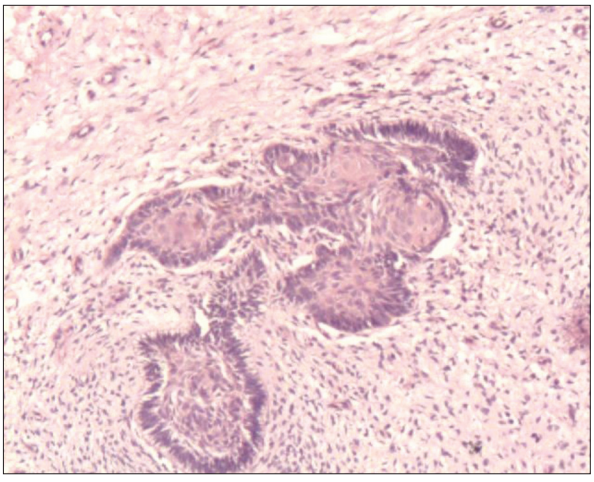

Figure 4: Photomicrograph showing tumor mass made up of epithelial and mesenchymal component of odontogenic origin (H and E, ×100)

Surgical excision of the lesion with curettage of surrounding bone was performed. The post-operative course was uneventful and the patient was discharged for further follow up. On follow up healing was uneventful and there were no signs of recurrence.

DISCUSSION

AF are reported to occur at an age ranging from 6 months to 42 years with an average age of 14.6-15.5 years.[6] The youngest patient reported by Mosby was a 7 week old infant. The reported sex predilection varies from no preference[7] to a male:female ratio that can vary from 2:1 to 4.4:3.[6] Most of these tumors occur in the mandible, with percentage varying from 83% to 90%.[3] The posterior portion of the dental arch is commonest location for most of the AF with the molar favored over the premolar location.[4] Very few cases are reported in the maxilla.

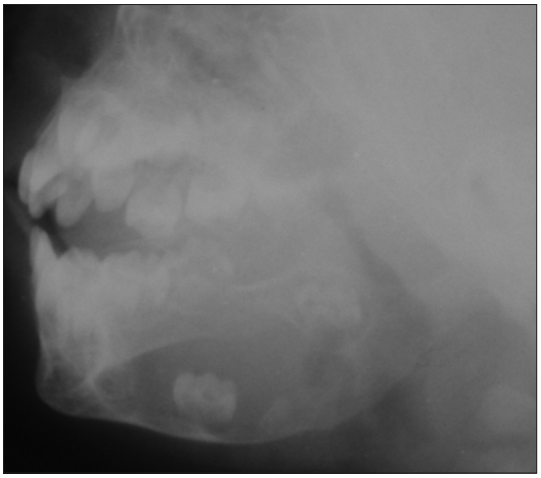

Figure 3: Lateral oblique view demonstrating the lesion

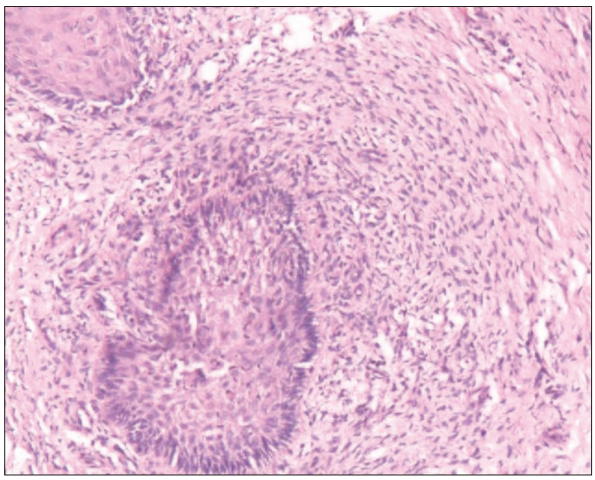

Figure 5: Photomicrograph showing epithelial component consisting of multiple, sharply defined strands or islands which are bordered at the periphery by a layer of tall columnar cells resembling ameloblasts and connective tissue showing primitive dental papilla like cells (H and E, ×200)

These lesions are usually slow growing, asymptomatic and are discovered incidentally (17% cases) or because of growth causing expansion (58% cases).[1,6] Pain, tenderness or mild swelling of the jaw may induce the patient to seek aid from the dentist. Tumors can vary in size from 1 cm to 8.5 cm and are reported to enlarge by extension as a solid mass.[4,7] They do not infiltrate and are not aggressive.[6]

Radiographically, AF usually appears as unilocular or multilocular radiolucency with well-defined and corticated borders in the premolar-molar area of the mandible. In some cases, the tumor may involve the ramus and extend forward to the premolar-molar area. Most of the lesions are associated with unerupted tooth as in our case. The cortical plates are thinned, expanded and intact.[5,8]

The differential diagnosis for AF includes intrabony cysts such as dentigerous cysts and odontogenic keratocyst, odontogenic neoplasms including ameloblastoma, myxoma, odontogenic fibroma and ameloblastic fibrosarcoma.

It is necessary to distinguish AF from ameloblastoma, ameloblastic fibrosarcoma since these two tumors can be locally invasive and have greater potential for recurrence than AF. Clinically AF occurs at a younger age than ameloblastoma. Radiographic examination however does not contribute to the differential diagnosis because all these tumors may appear as unilocular or multilocular or circumscribed lesions. Biopsy and histological examination however, will usually establish the diagnosis.[9]

AF are treated by enucleation and curettage of the surrounding bone and removal of the affected teeth.[9] Zallen *et al*., proposed microscopic examination of the surrounding bone at the site of AF to detect microscopic projections as seen in ameloblastoma.[7] Therefore, they advocate a more aggressive therapy that is modified block resection of the mandible with placement of an immediate autologous bone graft.

There are conflicting data in the literature on the recurrence rate of AF. Regezi *et al*., reported no recurrences in his series of 15 cases.[10] Tradhal *et al*. reported 10 recurrences in his series of 24 cases (approximately 44%).[6] In a review of the literature on recurrences of AF, Zallen *et al*., found a cumulative recurrence rate of 18.3%.[7] A large series of AF revealed that its recurrent rate at 10 years after the operation was approximately 70%.[2] In addition there have been a surprising number of cases reported of ameloblastic fibrosarcoma originating in some instances in recurrent AF.[11,12] Chen *et al*., found 14 out of 41 recurrent AF cases developed malignant changes.[2]

After evaluation of the literature, we believe that conservative removal of the initial AF lesion with modified block resection of any recurrences is a sound approach. Regardless of the form of treatment, patients with AF must be followed up for a long period to enable the early detection of possible recurrence or development of ameloblastic fibrosarcoma.

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How to cite this article: Munde AD, Karle RR, Kale UB. Ameloblastic fibroma in one-year-old girl. J Oral Maxillofac Pathol 2013;17:149.

Source of Support: Nil. **Conflict of Interest:** None declared.