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Case Report Open Access

Ameloblastic Fibro Odontoma Associated with Odontogenic Keratocyst – A Rare Case Report

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Abstract

Ameloblastic fibro odontoma is a rare benign odontogenic tumor that occurs in early decades of life. It shows male predilection and mandible is more affected when compared to maxilla. Clinically, it appears as a slow growing painless swelling and the patients may report with a complaint of failure of eruption of tooth. Radiographically, it appears as a well-defined unilocular or multilocular radiolucency with radio-opaque masses. The treatment of choice is surgical management like enucleation or resection. Based on the literature, the rate of recurrence was low. Ameloblastic Fibro Odontoma in a 10 year old female was reported in the department of oral and maxillofacial surgery. The present case has clinical interest because of its association with an odontogenic cyst. To our knowledge this is the first case report of Ameloblastic Fibro odontoma that has associated with a Odontogenic Keratocyst. The patient came with a swelling over her left mandible region. It was treated by enucleation under General anesthesia. Post operatively, the patient was followed up for 6 months and no signs of recurrence were noted. Mixed Odontogenic Tumor is a group of rare lesions that may delude the clinician to arrive at a many differential diagnosis. Any lesion which is painless and is associated with impacted tooth or odontoma is suspicious for Ameloblastic Fibro Odontoma.

Keywords: Ameloblastic fibro odontoma; Odontogenic; Enucleation; Resection; Odontogenic Keratocyst

Introduction

AmeloblasticFibro-Odontoma (AFO) has been defined by the WHO as a rare odontogenic tumor with the histopathological features of an ameloblastic fibroma (AF) in conjunction with the presence of dentin and enamel. It constitutes about 1 to 3% of all odontogenic tumours [1]. Neville et al has classified Ameloblastic fibro odontoma under 'mixed odontogenic tumor'[2]. World Health Organization in 2005 classifies Ameloblastic fibro odontoma under 'lesions with odontogenic epithelium with odontogenic ectomesenchyme, with or without hard tissue formation [3]. It is usually reported in 1st, 2nd and 3rd decade of life. However lesions during 4th decade of life were also reported [4]. This lesion shows male predilection and posterior mandible is most commonly involved area [1]. The lesion can be central or peripheral, central lesions being the most common type [5]. The present case reported was an ameloblastic fibroma associated with Odontogenic Keratocyst in a 10 year old female patient.

Case report

A 10 year old female patient came to the department of Oral and Maxillofacial Surgery with a chief complaint of a painless swelling over her left lower face region since 8 months. Past medical history and dental history were noncontributory. She gave no history of local trauma or infection.Extraorally, on examination an oval shaped well defined swelling approximately measuring about 4 x 6 cm was evident over the left lower third of the face along the lower border of the mandible. The skin overlying the lesion appeared normal (Figure 1). Mouth opening was normal.Intra orally the patient had mixed dentition. The swelling was extending from 34 regions in the anterior and posteriorly till the ramus.Vestibular obliteration was noted in relation to the lesion with a normal overlying mucosa. The teeth involved in the lesion were vital. On palpation, the swelling was hard in consistency.L esional aspiration was performed which showed straw colored fluid. No palpable lymph nodes were present. OPG showed well defined multilocular radiolucency extending from 34 region till the ramus region. Tooth like radio opaque mass was noted between 36 and 37 (Figure 2). Occlusal radiograph showed bicortical expansion. Based on the clinical and radiographic findings, the lesion was provisionally diagnosed as Odontogenic Keratocyst. The differential diagnosis included ameloblastoma, ameloblastic fibroma odontoma, calcifying odontogenic cyst or calcifying epithelial odontogenic tumor. Fine Needle Aspiration Cytology showed no

atypical cells. Incisional biopsy was also performed.Histologic examination showed 2-4 layers of non-keratinized lining epithelium. The epithelium connective tissue interface was flat and showed no rete pegs. Few clusters of round to oval epithelial cells with hyperchromatic nuclei was seen in connective tissue suggestive of odontogenic epithelial islands (Figure 3). On correlating the histopathologic feature with the clinical features of the lesion, final diagnosis of Odontogenic Keratocyst was arrived.

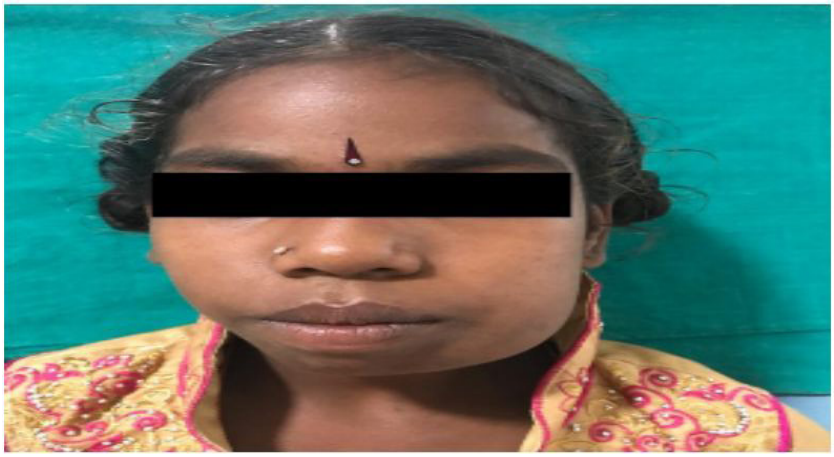

Figure 1: Preoperative extra oral view of the patient

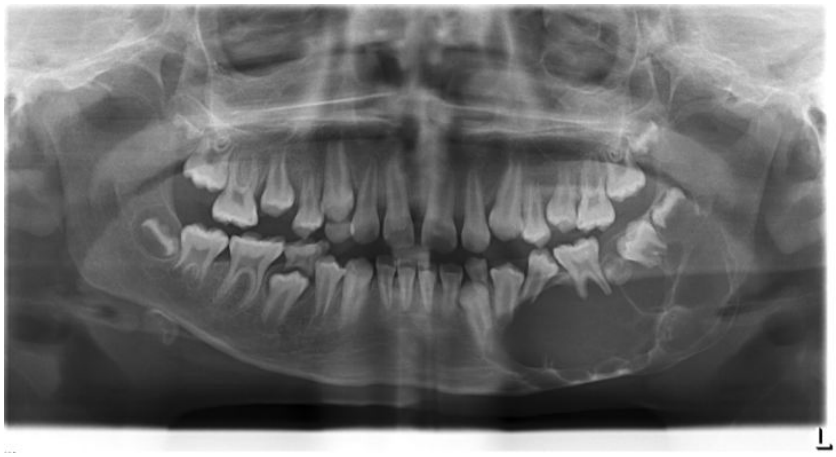

Figure 2: Radiographic view of the lesion

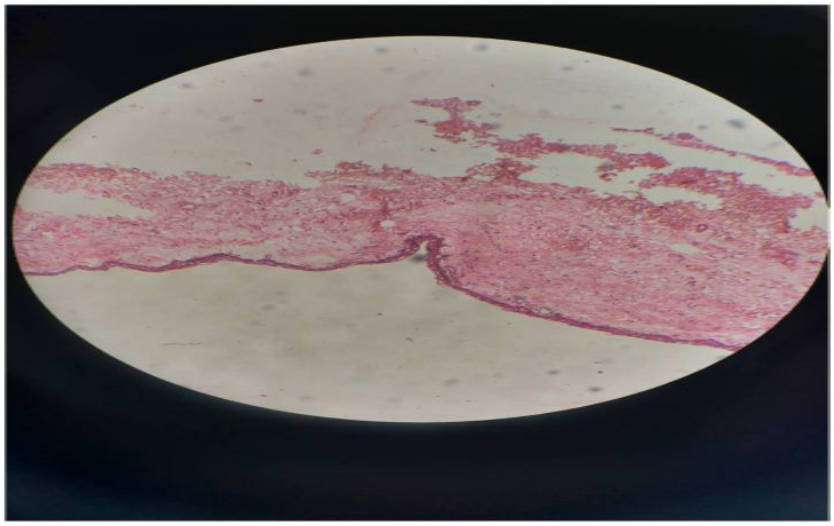

Figure 3: Histopathological view of incisionl biopsy

The planned management was nucleation of the lesion under general anesthesia. But the patient did not report for the next 3 months. Later she came with the complaint of increasing size of the swelling. On examination, the size of the lesion has increased to 5 x 7 cm. The patient was posted for nucleation under general anesthesia. The lesion was approached extra orally. No intraoperative or postoperative complications were encountered. Histopathologic examination showed lamellated bone with fibro collagenous

stroma and lined by squamous epithelium and area of primitive mesoderm with focal thin strands of odontogenic epithelium resembling dental lamina. Areas of granulation tissue, chronic inflammatory cells and multinucleated giant cells were seen which was suggestive of Ameloblastic fibro-odontoma (Figure 4). Patient was followed up for 6 months and showed no signs of reccurance (Figure 5). The radiographic examination showed good bone formation.

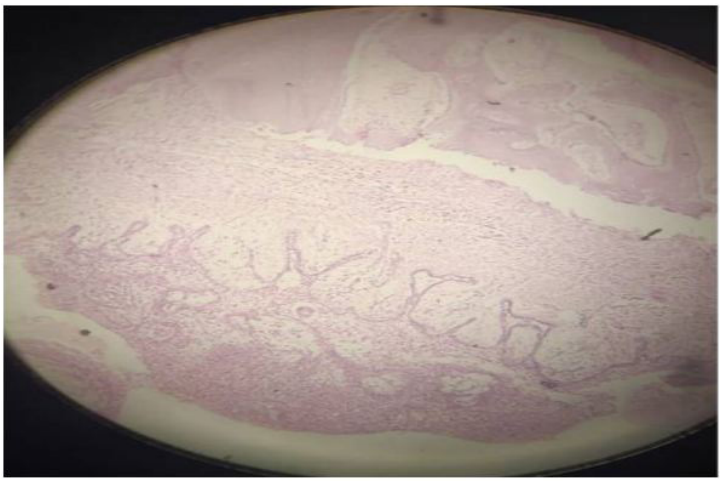

Figure 4: Histopathological view of excisional biopsy

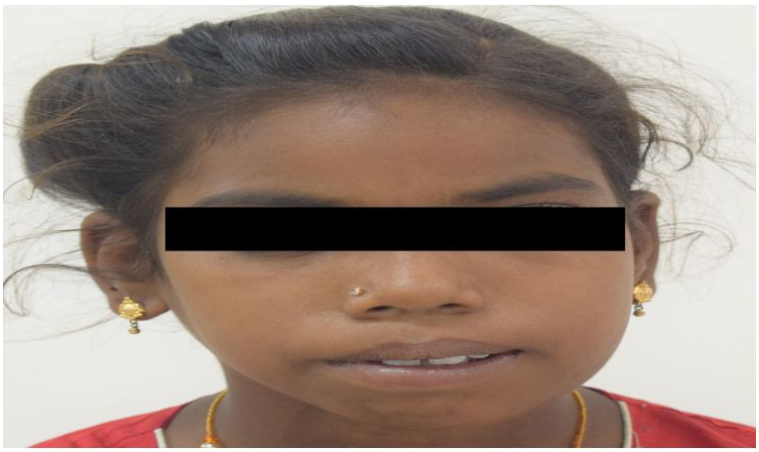

Figure 5: Follow up of the patient after 6 months

Discussion

Ameloblastic Fibro Odontoma is an immature complex odontome that represents hamartomatous rather than neoplastic lesion [6]. Clinically, the two main presentations are painless swelling and failure for tooth eruption [7] which holds good for the presented case. Thee presented case is a female patient which is contrast to the male predilection as given in the literature.

Radiographically, most of the reported lesions appear unilocular and only a very few cases were multilocular [8]. In the presented case, the radiographic appearance of the lesion was no way related to its pathognamic appearance which is well defined unilocular radiolucency. Almost all lesions were associated with complex odontome or the crown of an unerupted tooth [9] which in our case holds good.Histologically, it appears as primitve odontogenic epithelium resembling dental lamina. Mesenchymal components including fibrous connective tissue and fibroblasts will also be seen. Appearance of complex odontoma is typical. In the literature, 130 cases were documented out of which 11 cases were reported in India. Most of the cases were managed conservatively by enucleation and only a very few number of cases were managed by marginal resection [10]. Among the reported recurrent cases, the locularity of the lesions were compared and unilocular lesions were found to have higher recurrence [11]. In a case series study, 89 cases of benign odontogenic tumours were reviewed. It included Ameloblastic fibroma, ameloblastic odontoma and ameloblastic fibro odontoma. It was found the 22 case had undergone malignant transformation into Ameloblastic Fibro sarcoma . Out of this 22 cases, majority of the cases were Ameloblastic fibroma [12]. However the exact process of malignant transformation couldn't be found. It is noted that the transformation was followed by replacement of epithelial component by fibro sarcomatous component. Prognosis of Ameloblastic Fibrosarcoma is very poor. Half of the reported cases showed recurrence and most of the recurrence happened within the first 2 years.

Conclusion

Even though Ameloblastic Fibroodontoma exhibits a very subtle nature and can be treated by conservative surgery, its potential of malignant transformation should also be taken into consideration. Thus long term follow up for such cases are highly recommended.

References

1. Buchner A, Kaffe I, Vered M (2013) Clinical and Radiological Profile of Ameloblastic Fibro-Odontoma: an Update on an Uncommon Odontogenic Tumor Based on a Critical Analysis of 114 Cases. Head Neck Pathol 7: 54-63.

2. Neville BW, Damm DD, Allen CM, Chi AC (2015) Oral and maxillofacial pathology. Oral maxillofacial Pathol May 13.

3. Okui T, Ibaragi S, Shimo T, Takabatake K, Fujita M (2018) Ameloblastic fibro-odontoma in a 9-year-old boy. Stomatological Dis Sci 7: 4-8.

4. Mounir O , Ali BR , Messaoud BN , Chebbi K , Zakhama A , Hamdi H , Selmi J (2017) Ameloblastic Fibro-Odontoma of the Mandible: Uncommon Case Report. Sch J Dent Sci 4: 496-99.

5. Nayak B, Patankar S, Khot K, Shobha BV, Sharma G (2011) Ameloblastic fibro-odontoma of the maxilla: review of literature and report of a case. J of Indian Acad of Oral Med and Radiol 23: 442-44.

6. Shafer WG, Hine MK, Levy BM, Rajendran R, Sivapathasundharam B (1983) A textbook of oral pathology. Philadelphia: Saunders, USA.

7. Silva GC, Jham BC, Silva EC, Horta MC, Godinho SH, et al (2006) Ameloblastic fibro-odontoma. Oral Oncology Extra 42: 217-20.

8. Reis SR, de Freitas CE, do Espírito Santo AR (2007) Management of ameloblastic fibro-odontoma in a 6-year-old girl preserving the associated impacted permanent tooth. J of oral Sci 49: 331-5

9. Kirjavainen A, Tuovinen V, Sándor GK (2016) Large ameloblastic fibro-odontoma in a 7-year-old girl with analysis of 108 cases. Ann of maxillofacial Surg 6: 15-20.

10. Bernhoft CH, Bang G, Gilhuus-Moe O (1979) Ameloblastic fibro-odontoma. Int j of oral Surg 8: 241-4

11.Chrcanovic BR, Gomez RS (2017) Ameloblastic fibrodentinoma and ameloblastic fibro-odontoma: an updated systematic review of cases reported in the literature. J of Oral and Maxillofacial Surg 75: 1425-37.

12. Friedrich RE, Siegert J, Donath K, Jäkel KT (2001) Recurrent ameloblastic fibro-odontoma in a 10-year-old boy. J of oral and maxillofacial Surg 59: 1362-6.

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