Acta Scientific Otolaryngology

Volume 2 Issue 2 February 2020

Case Report

Ameloblastic Fibroma of the Maxilla - A Rare Case Report

Amrapali Keny Pawar[1]* and Vishakha Rane[2]

[1]Honorary, ENT Surgeon, V. N. Desai Municipal Hospital, Mumbai, India

[2]House Officer, ENT Surgeon, V. N. Desai Municipal Hospital, Mumbai, India

***Corresponding Author:** Amrapali Keny Pawar, Honorary, ENT Surgeon, V.N. Desai Municipal Hospital, Mumbai, India.

Received: January 23, 2020

Published: January 30, 2020

Abstract

Ameloblastic fibroma is a benign odontogenic tumor. These are extremely rare benign neoplasm affecting usually young children. It is a mixed odontogenic, non-encapsulated tumor. Most common site of manifestation being the mandibular premolar-molar region.

Here, we report a rare case of 60 years old male with a painless expansile swelling arising from the right maxilla which was subsequently diagnosed as ameloblastic fibroma.

Keywords: Rare; Benign; Mixed Tumor; Ameloblastic Fibroma; Maxilla; Odontogenic Tumor

Introduction

Ameloblastic fibroma is a rare, mixed, non-encapsulated, slow growing, benign odontogenic tumor. It is composed of mesenchymal tissue resembling dental papillae and islands of odontogenic epithelium resembling the dental lamina [1]. It is an extremely rare tumor accounting for about 2% of the odontogenic tumors [2,4]. They tend to develop 'de novo', without any apparent etiological factor. They are usually asymptomatic and young children less than 20 years are usually affected [3] with the most common site of manifestation being the mandibular premolar-molar region. Males are more commonly affected than females, ratio being 1.4:1 Most cases of ameloblastic fibromas are encountered as incidental findings [5,6].

Radiographically they usually present with well-defined unilocular or multi-locular radioluscencies but it may vary It is treated with surgical enucleation with close follow-up for recurrences [7].

Case Report

A 60years old male patient visited ENT OPD with chief complaints of swelling over right cheek since 6 years. The swelling was painless, progressive and gradually increasing. There was no difficulty in mastication, no history of difficulty in deglutition, no history of paraesthesia or fever. No significant past medical, surgical or family history. No history of any addictions.

On clinical examination there was facial asymmetry with well circumscribed swelling on the right cheek, measuring about 4x5cms in size, oval in shape, extending superiorly to the right zygomatic arch, medially up to the ala of right nostril, with no secondary changes over the skin, no visible pulsations, no cervical lymphadenopathy. On palpation, the swelling was nontender, non-mobile, no local rise of temperature, firm to bony hard in consistency (Figure 1,2). On intraoral examination there was diffuse swelling over the right upper lateral incisors, premolar, molars. The overlying mucosa was intact and healthy. No displacement of the hard palate (Figure 3,4).

Figure 1: Pre Operative picture of the patient showing lateral extension of the tumor.

Figure 2: Pre Operative picture of the patient showing asymmetric face.

A computed tomography was done which revealed an expansile osteolytic lesion in the right maxillary alveolous. The lesion shows multiple cortical breaches, multiple calcific and a few cystic foci. There was no erosion of the surrounding bone, no extension within

Figure 3 and 4: Intra Oral picture of the tumor.

the maxillary sinus. It extends intraorally to right side of upper gingivobuccal sulci and buccal pad of fat (Figure 5,6).

Figure 5: CT Image AP.

Figure 6: CT Image Lateral.

Based on the clinical and radiographic findings a provisional diagnosis of benign odontogenic tumor was made.

A surgical enucleation of the lesion, along with curettage of the adjacent maxillary bone and removal of teeth was performed under general anesthesia.

The surgical specimen was sent for histopathological examination. Macroscopically the specimen measured about 4 x 4.5 cms in greatest dimensions with smooth surface on one side and rough on the other side, was firm in consistency with few soft cystic areas (Figure 7,8,9). Microscopically the lesion revealed non-encapsulated neoplasm composed of ameloblastic epithelium embedded in a fibrous stroma with central foci of keratinisation in few islands suggestive of ameloblastic fibroma.

Figure 7 and 8: Post Operative Specimen.

Figure 9: Post Operative Day 2 picture of the patient.

Discussion

Ameloblastic fibromas were first described by Kruse in 1890 According to WHO (1992), ameloblastic fibroma is clearly a neoplasm of odontogenic origin with an epithelial and ectomesenchymal component.

It is histologically a biphasic tumor (true-mixed neoplasm) containing epithelial and ectomesenchymal components. It is predominantly located over unerupted molars in young patients. The male: female ratio is 1.4:1 according to Reichart and Philipsen. Mean age is 14.8 years, with a wide age range of 0.5 to 62 years. The youngest case reported was in a seven weeks old infant [12]. It is a slow growing tumor commonly located in the posterior mandible [9]. Incidence of maxillary ameloblastic fibromas is extremely rare. The case reported by us is an extremely rare case with maxillary ameloblastic fibroma in a 60 year old male patient.

Most of the cases of ameloblastic fibromas are incidental findings, very few cases may come with presentation of AF as a unilateral painless slow growing swelling.

The radiographic features are variable, ranging from well circumscribed small lucent unilocular lesion to expansile multiloculated lesion, with well-defined borders and sclerotic margins [10,11].

Differential diagnosis of Ameloblastic fibroma may include ameloblastoma, odontogenic keratocyst, odontogenic myxoma, chondroid tumors.

Histological differentials include its malignant counterparts ameloblastic fibrosarcoma, ameloblastic fibrodentinoma, ameloblastic fibro-odontinoma.

Ameloblastic fibroma lacks dental hard tissue like enamel or dentin which is present in ameloblastic fibrodentinoma or ameloblastic fibro-odontinoma as well as ameloblastoma.

Ameloblastic fibrosarcoma is composed of benign epithelium and malignant mesenchymal tissue comprising marked cellularity and high mitotic figures. These characteristic findings were absent in our case, thus giving the diagnosis of ameloblastic fibroma.

Ameloblastoma is histologically composed with epithelium of the tooth germ, epithelium of enamel, stratified squamous epithelium of odontogenic cysts. It is more aggressive than ameloblastic fibroma. Our patient's specimen showed proliferation of both odontogenic epithelium and mesenchymal tissue embedded in fibrous stroma, without the formation of enamel or dentin. Thus confirming our diagnosis of ameloblastic firboma.

The management of ameloblastic fibroma is enucleation of the tumor with preservation of the surrounding vital structures and close watch for recurrence.

Ameloblastic fibromas show variable rate of recurrence with few cases turning to malignant ameloblastic fibrosarcoma. The review of literature for recurrence rate shows that Trodahl., *et al.* found the rate to be 43.5% [13], on the other hand Zallen., *et al.* found it to be 18.3% [14]. The reason for discrepancy in recurrence rates is uncertain, suggesting the probable cause of recurrence being incomplete removal or presence of satellite tumors at the edges of the lesion. It is thought that around one-third of ameloblastic fibrosarcomas develop as a result of malignant transformation of an ameloblastic fibroma [15]. Thus close follow-up is necessary postoperatively.

Conclusion

This case demonstrates that ameloblastic fibromas can be managed by complete enucleation on the lesion with preservation of the surrounding vital structures. However a careful follow-up of the patients is necessary owing to the variable recurrence rates of ameloblastic fibromas and their potential to turn into malignant ameloblastic fibrosarcoma, especially in older age groups.

Bibliography

1. RH Kramer., *et al*. "The WHO histological typing of odontogenic tumors". *Cancer* 70 (1992): 2988-2994.
2. Regezi JA., *et al*. "Odontogenic tumors: analysis of 706 cases". *Oral Surgery* 36 (1978): 771-778.
3. PJ Slootweg. "An analysis of the interrelationship of the mixed odontogenic tumors-ameloblastic fibroma, ameloblastic fibro-odontoma, and the odontomas". *Oral Surgery Oral Medicine and Oral Pathology* 51.3 (1981): 266-276.
4. DM Cohen and I Bhatttacharyya. "Ameloblastic fibroma, ameloblastic fibro-odontoma, and odontoma". *Oral and Maxilllofacial Surgery Clinics of North America* 16.3 (2004): 375-384.
5. Y Chen., *et al*. "Ameloblastic fibroma: a review of published studies with special reference to its nature and biological behavior". *Oral Oncology* 43.10 (2007): 960-969.
6. L Barnes., *et al*. Pathology and Genetics of Tumors of the Head and Neck: World Health Organization Classification of Tumors: International Histological Classification of Tumors, IARC Press, Lyon, France, 3rd edition (2005).
7. RS Kulkarni., *et al*. "Recurrent ameloblastic fibroma: report of a rare case". *Case Reports in Dentistry* (2013).
8. Reichart PA and Philipsen HP. Odontogenic Tumors and Allied Lesions. Hanover, Germany. Quintessence (2004).
9. Bell RA., *et al*. "Ameloblastic fibroma: A case report". *Pediatric Dentistry* (1982).
10. LR Cahn and T Blum. "Ameloblastic odontoma: case report critically analysed". *Journal of Oral Surgery* 10 (1952): 169-170.
11. A Buchner and M Vered. "Ameloblastic fibroma: a stage in the development of a hamartomatous odontoma or a true neoplasm? Critical analysis of 162 previously reported cases plus 10 new cases". *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology* 116.5 (2013): 598-606.
12. EL Mosby., *et al*. "Ameloblastic fibroma in a 7 week-old infant: a case report and review of the literature". *Journal of Oral and Maxillofacial Surgery* 56.3 (1998): 368-372.
13. Trodahl JN. "Ameloblastic fibroma. A survey of cases from the armed forces institute of pathology". *Oral Surgery, Oral Medicine, Oral Pathology* 33 (1972): 547-58.
14. Zallen RD., *et al*. "Ameloblastic fibroma". *Journal of Oral Maxillofacial Surgery* 40 (1982): 513-517.
15. A Kousar., *et al*. "Rapid sarcomatous transformation of an ameloblastic fibromabof the mandible: case report and literature review". *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology and Endodontics* 108.3 (2009): e80-e85.