Ameloblastic Fibro-Odontoma of the Maxilla: Review of Literature and Report of a Case

Barakha Nayak, Sangeeta Patanker, Komal Khot, Shobha BV, Gopal Sharma

1Senior Lecturer, Department of Oral and Maxillofacial Pathology, Maitri College of Dentistry and Research Centre, Durg, Chhattisgarh, India
2Professor and Head, Department of Oral Pathology, YMT Dental College and Hospital, Navi Mumbai, Maharashtra, India
3Professor, Department of Oral Medicine, YMT Dental College and Hospital, Navi Mumbai, Maharashtra, India
4Professor, Department of Oral Pathology, Maitri College of Dentistry and Research Centre, Durg, Chhattisgarh, India

Correspondence: Barakha Nayak, Senior Lecturer, Department of Oral and Maxillofacial Pathology, Maitri College of Dentistry and Research Centre, Durg, Chhattisgarh, India, e-mail: brkh_verma@yahoo.co.in

ABSTRACT

Ameloblastic fibro-odontoma (AFO) is a rare benign mixed odontogenic tumor. It is a slow-growing generally asymptomatic lesion and more prevalent in children and adolescents. This article presents interesting case of ameloblastic fibro-odontoma involving anterior maxilla of 15-year-old male patient. Occlusal radiograph showed a large, well-demarcated radiolucency with radiopaque areas and miniature teeth like structures, radiographical presentation of this case was suggestive of compound odontoma. Histopathologically, the lesion was diagnosed as ameloblastic fibro-odontoma. This article discusses about clinical, radiographical, histopathological aspect of ameloblastic fibro-odontoma with review of literature.

Keywords: Ameloblastic fibro-odontoma (AFO), Odontogenic tumors.

INTRODUCTION

Odontogenic tumors are a heterogeneous group of diseases ranging from hamartomas to benign and malignant neoplasms. Odontogenic tumors arise from odontogenic epithelium, ectomesenchyme and mesenchymal tissues. Recent studies assessing large numbers of cases have shown that these tumors constitute 0.84 to 1.78% of the histopathological findings of renowned oral pathology departments. According to the World Health Organization, ameloblastic fibro-odontoma (AFO) is a tumor with histological features similar to those of ameloblastic fibroma (AF), but with inductive changes that lead to the formation of dentin or enamel. Among the odontogenic tumors, incidence of AFO varies from 0.3 to 1.7%, reaching 4.6% when only the cases in children are considered. Most cases are diagnosed in the patient’s first two decades of life, between the ages of nine and eleven years on average. There is a slightly higher incidence of AFO in males, most commonly seen in the posterior mandible area. AFO is generally an asymptomatic, slow-growing tumor, commonly associated with an unerupted tooth. Radiographically, ameloblastic fibro-odontoma presents as a well-demarcated radiolucency containing radiopaque areas. Conservative surgical excision is the treatment of choice and the lesions does not tend to recur. Histopathologically, islands, strands and cords of odontogenic epithelium immersed in embryonic connective tissue that mimics primitive dental pulp with formation of osteodontin and enamel are seen.

CASE REPORT

A 15-year-old male patient reported to YMT Dental College with the chief complaint of missing upper front teeth and swelling since 2 months. The swelling had started as a small bulging and had gradually increased to the present size. Patient gave the history of trauma 9-year back in the same region and his medical history was unremarkable. Extraoral examination revealed mild facial asymmetry, with swelling of the left side of the maxilla which was asymptomatic and covered with healthy skin of normal color. Intraoral examination revealed a swelling in anterior palatal area, extending from the left deciduous lateral incisor to right permanent central incisor covered with normal mucosa, 3 × 4 cm in size, over-retained deciduous lateral incisor and transposition of permanent lateral incisor of left side were seen, swelling was nontender and hard to palpation (Fig. 1). In occlusal radiograph circumscribed radiolucent lesion containing radiopaque masses of varying sizes and shapes were seen along with impacted 21, 23 and over-retained 62 (Fig. 2). Based on the clinical and radiographical findings, a provisional diagnosis of compound odontoma was given. The differential diagnosis should include lesions with mixed radiographic patterns, such as, immature complex odontoma and possibly adenomatoid odontogenic tumor.

Under general anesthesia curettage of the lesion and extraction of 62 were done. Postoperative recovery was uneventful and patient was advised a follow-up examination after 1 month. Excised tissue grossly showed a mixed lesion...
some part made up of soft tissue and other part composed of calcified materials, rudimentary teeth like structures were also seen (Fig. 3).

Histopathological examination revealed immature connective tissue stroma with young fibroblasts and delicate collagen fibers containing cords, strands and islands of pre-ameloblast like odontogenic epithelium. At places these islands have peripheral halo of hyalinization which is suggestive of inductive changes. At places abundant dentin-like tissue enclosing pulp-like tissue and osteoid like tissue are seen (Fig. 4). The present case was histopathologically diagnosed as ameloblastic fibro-odontoma associated with compound odontoma.

DISCUSSION

Hooker in 1967 suggested the present terminology ameloblastic fibro-odontoma as an entity until then many similar odontogenic tumors were categorized as ameloblastic fibroma, ameloblastic odontoma and cyst adenoma. It is a rare benign mixed odontogenic tumor composed of ameloblastic fibromas on one hand and complex odontoma on the other. Controversy exists regarding the histogenesis of mixed odontogenic tumors. Cahn and Blum (1952) postulated that ameloblastic fibroma, the histologically least differentiated tumor, develops first into a moderately differentiated form, ameloblastic fibro-odontoma and eventually into complex odontoma. However, the concept that these lesions represent a continuum of differentiation is not widely accepted, and others feel that they are separate pathologic entities.

There has been a lot of discussion in the literature regarding whether AFO is a neoplasm or hamartoma. HP Philipsen and Reichart (1997) suggested a hypothesis regarding the pathogenesis and relationship between the ‘mixed odontogenic tumors’ and the odontomas. Most mixed odontogenic tumors are considered to be hamartomatous and are part of a developing complex odontoma line. AF or AFD is the first step in the development of a complex odontoma. These tumors can develop further into the second stage called AFO. The final stage is the
fully mineralized complex odontoma. The compound odontoma
is not an alternative final stage of the complex odontoma but
rather as a malformation with a high degree of histomorpho-
logical differentiation, pathogenetically closely related to the
process producing hyperodontia, ‘multiple schizodontia’ or
locally conditioned hyperactivity of the dental lamina.5 The
diagnosis of present case ameloblastic fibro-odontoma
associated with compound odontoma was supported by this
theory.

According to the World Health Organization, the distinction
between complex and compound odontomas is based on the
arrangement of the dental hard tissues found in the lesions. In
complex odontomas, these tissues are found in a more or less
disorderly pattern, whereas in compound odontoma denticles
are found. However, some authors have suggested that both
types of odontoma are pathogenetically different. A complex
odontoma, more commonly found in the posterior mandible,
like AFO, could be the end-stage of a hamartomatous lesion.
On the other hand, a compound odontoma seldom has any
clinical relation with AFO and its occurrence could be the result
of local hyperactivity of the dental lamina. According to Chen
et al, the presence of disorganized odontogenic epithelium and
ectomesenchyme, associated with irregular formation of dentin
and enamel make it unlikely that structures similar to teeth
formed in AFO.6 Nevertheless, in the present case reported here,
rudimentary teeth like structure were quite apparent.

AFO is treated by curettage as it does not appear to locally
invade the bone.7 All cases of AFO should not be considered
hamartomatous, since there are cases that show true neoplastic
behavior,7 and because there is evident existence of a malignant
variant of the disease.8,9 Thus one may suppose, it is possible
that different lesions are being described under the same term
of AFO, with some being hamartomatous in nature and others
being a true de novo neoplasm.6

CONCLUSION

The unique case of ameloblastic fibro-odontoma associated with
compound odontoma was reported in accordance with the
literature. There was no recurrence noted clinically and
radiographically till date. Long-term follow-up is advised in
management of ameloblastic fibro-odontoma.

REFERENCES

Organization classification of tumours. Pathology and genetics
2. Guerrisi M, Piloni M, Keszler A. Odontogenic tumors in children
and adolescents. A 15-year retrospective study in Argentina.
3. Shafer WG, Hine MK, Levy BM. A textbook of oral pathology
4. Slootweg PJ. Analysis of the interrelationship of mixed
odontogenic tumors–ameloblastic fibroma, ameloblastic fibro-
5. Philipsen HP, Reichart RA, Pratorius F. Mixed odontogenic
tumors and odontomas: Considerations on interrelationship:
Review of the literature and presentation of 134 new cases of
6. Chen Y, Tie/Jun L, Yan G, Shi-Feng Y. Ameloblastic fibroma
and related lesions: A clinicopathologic study with reference to
their nature and interrelationship. J Oral Pathol Med 2005;34:
588-95.
7. Miller AS, Lopez CF, Pullon PA, Elzay RP. Ameloblastic fibro-
8. Howell RM, Burkes EJ. Malignant transformation of
ameloblastic fibro-odontoma to ameloblastic fibrosarcoma. Oral
of the mandible: Report of two cases and review of the literature.