Ameloblastic Fibro-Odontoma: A Case Report

Satya Ranjan Misa1, Pavitra Baskaran2, G Maragathavalli3, R Mithra2, N M Praveena4

1Reader, Department of Oral Medicine and Radiology, Institute of Dental Sciences, Bhubaneshwar, Odisha, India, 2Reader, Department of Oral Medicine and Radiology, Meenakshi Ammal Dental College, Chennai, Tamil Nadu, India, 3Professor, Department of Oral Medicine and Radiology, Saveetha Dental College, Chennai, Tamil Nadu, India, 4Reader, Department of Oral Medicine and Radiology, Thai Moogambigai Dental College, Chennai, Tamil Nadu, India

Ameloblastic fibro-odontoma (AFO) is an uncommon benign odontogenic tumor with histologic features similar to that of ameloblastic fibroma, but with inductive changes that lead to the formation of enamel and dentin. Among the odontogenic tumors, the incidence varies from 0.3% to 1.7%, reaching 4.6% when only the cases of children are considered. AFO presents as a painless swelling mostly in the posterior portion of the maxilla or mandible. Radiographs show the radiolucent area containing various amounts of radiopaque material of irregular size and form. This case report describes AFO affecting the 21-year-old woman. The lesion was surgically excised, and no recurrence was observed on follow-up.

Keywords: Ameloblastic fibro-odontoma, Fibro-odontoma, Odontogenic tumor

INTRODUCTION

Odontogenic tumors are a heterogenous group of diseases ranging from hamartomas to benign and malignant neoplasms.1 Ameloblastic fibro-odontoma (AFO) is a rare benign, slow growing, expansile epithelial odontogenic tumor with odontogenic mesenchyme. The term AFO was first used by Hooker in 1967.2 AFO was originally termed as ameloblastic odontomas but in 1971, World Health Organization (WHO) suggested that this term is inappropriate since it encompasses two types of odontogenic tumors that are different in their histologic and biologic behavior.3 It is defined by WHO as a neoplasm composed of proliferating odontogenic epithelium embedded in a cellular ectomesenchymal tissue that resembles dental papilla, and with varying degrees of inductive change and dental hard tissue formation.4

A majority of AFO is intraosseous and is associated with unerupted teeth. It is reported that approximately 80% of the lesions were located in the posterior area of the jaws, and most (58%) were in the posterior mandible.5 54% of AFO are found in the posterior mandible.6 Radiographs show a well-defined radiolucent area containing various amounts of radiopaque material of irregular size and form.7 This report describes an AFO in a 21-year-old woman.

CASE REPORT

A 21 years female patient reported to the dental hospital with a swelling in the right side of her face for 2 weeks. History revealed that the swelling was slowly growing in size and was painless. The medical, personal, and family history was non-contributory.

A diffuse swelling was seen in the right body of mandible region, with ill-defined margins (Figure 1). The swelling was soft in consistency. Intraorally the swelling was observed obliterating the buccal sulcus in 46, 47 regions. It was fluctuant, non-tender, and fixed to the underlying bone. Correlating the history and clinical findings, a clinical diagnosis of the odontogenic cyst was made.

Following the clinical diagnosis, patient was subjected to the following investigations. Periapical radiograph was taken which revealed a well-defined, unilocular, inter-radicular radiolucency with a hyperostotic border was seen between the distal root of 46 and mesial root of 47 (Figure 2). Mandibular right lateral occlusal radiograph showed the expansion of the lingual cortex (Figure 3). Panoramic radiograph revealed a well-defined, ovoid, unilocular, inter-radicular radiolucency with a hyperostotic border measuring 1 cm in greatest diameter (Figure 4). Correlating the clinical and radiographic findings, a radiologic diagnosis of lateral periodontal cyst was made.

Corresponding Author:
Dr. Pavitra Baskaran, Department of Oral Medicine and Radiology, Meenakshi Ammal Dental College, Chennai - 600 082, Tamil Nadu, India.
E-mail: pavitrar misdemeanors@gmail.com
Aspiration of the cystic content was made, and a straw colored fluid was obtained on aspiration. Excisional biopsy was performed and Hematoxylin–Eosin (H and E) stained section revealed narrow cords and small discrete islands of odontogenic epithelium with loose primitive connective tissue resembling dental papilla (Figure 5a). There were areas showing primitive mesenchyme with mild basophilia and angular fibroblasts (Figure 5b) and odontogenic islands with eosinophilic rimming resembling dentinoid (Figure 5c). Odontogenic islands show peripheral columnar cells resembling ameloblasts, which surround a mass of loosely arranged epithelial cells resembling stellate reticulum (Figure 5d). All the above histologic features were suggestive of ameloblastic fibrodontoma.

The lesion was treated by complete surgical excision. The patient was followed up for 6 months without any signs of recurrence.

**DISCUSSION**

AFO is a rare odontogenic tumor of childhood and adolescence. It is seen in young patients, with the mean age of 11.5 years. In general, it is seen in the first and second decades of life, which might also be a characteristic of the lesion. However, AFO may also occur at advanced ages.
ages. Previously, Philipsen et al. declared that the mean age of AFO cases falls when compared with the ameloblastic fibroma (AF) and AF-dentinoma, supporting the suggestion that age is a critical feature in AFO diagnosis. The frequency of occurrence of AFO is about 1-3% considering all odontogenic tumors. The two main complaints are swelling and failure of tooth eruption. Clinically, it presents as a painless swelling of the affected area and is usually seen in the posterior maxilla or mandible as seen in our case.

Radiographically, most authors state that it presents as a unilocular or multilocular lesion. AFO appears as a radiolucent area that consists of the variable amount of calcified material. The center shows a radiopacity irregular in shape and density or may present as homogenus rounded calcified mass. The ratio of radiopaque to radiolucent areas differs from one lesion to another, sometimes the mineralized elements in the tumor predominate and the lesion may be radiographically similar to that of an odontoma.

A diagnosis of AFO is given when the lesion presents with typical age, location, and radiographic pattern. Differential diagnosis of AFO based on mixed radiographic patterns includes lesions such as calcifying epithelial odontogenic tumor, calcifying odontogenic cysts, immature complex odontoma, and adenomatoid odontogenic tumor.

Histologically, AFO is composed of strands, cords, and islands of odontogenic epithelium embedded in a cell rich, primitive ectomesenchyme that resembles the dental papilla. Dentin and enamel matrix are also seen. Our case reported here is consistent with the above findings.

Histopathological differential diagnosis also includes AF, ameloblastic fibro-dentinoma. However, AF and AFO are believed to be stages of complex odontoma formation. Cahn and Blum postulated that AF develops first into moderately differentiated form, following AFO, and eventually into a complex odontoma. The concept of a continuum of differentiation is not widely accepted, with other researchers suggesting that they are separate pathologic entities. AFO exists as a distinct entity, but histologically it is indistinguishable from immature complex odontoma. However, AFO differs significantly from the hamartomatous odontoma by having a greater potential for growth and causing considerable deformity, bone destruction.

It is not possible to distinguish hamartoma from neoplasm based on histological features alone. Philipsen and Reichart (1997) suggested a hypothesis regarding pathogenesis. Most mixed odontogenic tumors are considered to be hamartomatous in nature and are a part of compound odontoma line. AF or fibro-dentinoma is the first step in developing compound odontome. These tumors can develop further in the second stage of AFO. The final stage is the fully mineralized complex odontoma with high degree of histomorphologic differentiation.

The presence of dentin and enamel matrix is the feature that separates the AFO from AF. The amount of mineralized products of odontogenesis may vary from being easily seen on grossing to require serial sectioning to identify dentin and enamel matrix microscopically.

AFO shows dental structures resembling dentine, but also because it contains enamel-like tissues. Thus, the formation of AFO might be based on enamel matrix production, which is one of the most important features of the lesion differentiating it from ameloblastic fibro-dentinoma.

The treatment of AFO is by conservative surgical excision. However, if the lesions are larger, it may require surgical resection of either partial maxillectomy or partial mandibulectomy.

Recurrence of AFO is not common. Some authors have reported recurrence in cases where the impacted or retained teeth have been preserved. The recurrence is governed by the degree of involvement of impacted tooth by the tumor and the difficulty to completely remove the lesion. In the present case, no recurrence was reported after the surgical excision that was followed up after 6 months.

CONCLUSION

AFO is a rare benign odontogenic tumor that emphasizes the importance of radiologic examination in early detection. Although rare, AFO should be considered in the differential diagnosis of intra-oral radioluencies that contain radiopaque material.

REFERENCES

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