Adenomatoid Odontogenic Tumor, An Uncommon Cause of Facial Swelling

Parveen Akhter Lone[1], Ambica Sudan[2], Jehangir Ahmed[2], Tahir[2]

ABSTRACT

Introduction: Adenomatoid odontogenic tumor (AOT) is an un common tumor of odontogenic origin constituting only 3 % of all odontogenic tumors. Adenomatoid odontogenic tumor (AOT) is a slow growing, benign epithelial tumor that presents with absence of teeth and facial deformity in very large lesions. The aim of this study was to report the rare and uncommon cause of facial swelling the follicular and extra follicular AOT, in young females and to document the clinical presentation and treatment protocols for AOT of jaws.

Material and Methods: The prospective study was conducted in OMFS department. All the patients with swelling of maxilla/ facial deformity were included in the study. The duration of this study was from January 2010 – december 2015. Eight patients presented with the features of AOT. After detailed history, examination, OPG and CT scan provisional diagnosis was made. After informed consent incisional biopsy was done in all the cases under local anesthesia to confirm the diagnosis

Results: In 5 years period eight cases were diagnosed AOT, five were females and three males. Age ranged between 12-21 years. Maxilla was more involved more than mandible. Enucleation / surgical excision was done in all cases.

Conclusion: Management of the AOT should be done in the initial days by excising it in toto. All such cases should be reported in the literature for the betterment of mankind.

Keywords: Impacted teeth, True neoplasm, Facial deformity, AOT

INTRODUCTION

Adenomatoid odontogenic tumor (AOT) is a slow growing, benign, epithelial tumor that presents with clinical signs of the absence of teeth and, in the case of very large lesions, with deformity. Adenomatoid odontogenic tumor is usually seen during the second and third decade of life. The incidence is higher in females tham males.[1] Adenomatoid odontogenic tumor is usually located in the anterior region of the maxilla with in which an impacted tooth exists.[2] It ia an un common tumor and constitutes only 3 to 7 % of all odontogenic tumors.[3] It was first reported by Harbit[4] in 1915 as cystic ademantoma. Phillips and Brin[5] proposed the widely accepted currently used name Adenomatoid odontogenic tumor (AOT) a term first adopted by WHO in 1972 in world health organization classification of odontogenic tumors. WHO in the latest edition of classification of odontogenic tumors in 2005, classified AOT in fisrt group of tumors (odontogenic epithelium without ectomesenchyme) instead of second group odontogenic epithelium with ectomesenchyme.[6] AOT is characterized histologically by formation of duct like structures with ameloid type deposits.

Regarding its pathogenesis, the lesion originates from odontogenic epithelium(enamel organ or dental lamina remnants) with inductive influence on odontogenic ecto mesenchyme and consequent production of dentinoid material. Radiographicaly, the tumour usually appears as a well circumscribed, unilocular radiolucency with scelerotic areas which may be associated with an unerupted tooth, most often a canine.[7]

Tumor appears as intra oral –extra oral swelling of maxilla and is sometimes called as “two thirds tumor” because it occurs in maxilla in 2/3 cases, around 2/3 cases in young females, 2/3 cases associated with un erupted teeth and 2/3 affacted teeth are canines.[8] The lesion may contain only the crown of the tooth or whole tooth. 'It usually occurs in the anterior region of the maxilla[1]

Topographically AOT occurs in Peripheral and central variant, the latter type is further divided into follicular (with embedded tooth) and extra-follicular (without an embedded tooth) types. The central variant accounts for 97.2% and from which 73.0% of its type are follicular. The follicular variant (M;f ratio 1-1.9) is three times as frequent as extra-follicular type. The follicular variant is diagnosed earlier in life (mean17 yrs) than extra follicular type (mean 24yrs):53.1%of all variants occur within the teen years (13-19yrs). A study has shown that follicular AOT was associated with one embedded tooth in 93.2%.[9] Maxillary permanent canines account for 41.7% and all four canines for 60.1% of AOT-associated embedded teeth. The follicular variant of AOT is thought to be originated from the reduced enamel epithelium of the dental follicle. The origin of extra follicular remnants remains less clear. The available evidence seems indicate that some extra follicular AOTs might arise as secondary phenomenon within the pre-existing odontogenic cysts or cystic tumors.[10]

The aim of this study was to report the rare and uncommon cause of facial swelling the follicular and extra follicular AOT, in young females and to document the clinical presentation and treatment protocols for AOT of jaws

MATERIAL AND METOHDS

The prospective study was conducted in OMFS department. All the eight patients with swelling of maxilla/ facial deformity were included in the study. The duration of this study was from January 2010 – December 2015. Nine patients presented with the features of AOT like painless swelling of face, gingival, missing teeth. After detailed history, examination, OPG and CT

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scan provisional diagnosis was made. After informed consent incisional biopsy was done in all the cases under local anesthesia to confirm the diagnosis. All the cases were treated with wide surgical excision under GA, patients were discharged after sutures removal after seven days and healing was uneventful. No recurrence was reported (Figures1-4).

The histopathological findings revealed a benign encapsulated mass composed of whorls, sheets and strands of epithelial cells, along with microcysts and ducts lined by cuboidal to tall columnar cells. The microcystic contain an eosinophilic material. Sheets of epithelial cells showed the foci of amorphous material, areas of haemorrhage, dilated and congested blood vessels. It was diagnosed as an Adenomatoid odontogenic tumor.

RESULTS

Eight cases of AOT were recorded during the study. The age range was 12 -21 years and mean age was found 18 years. Lesion was seen more in females (70%) than males (5 females and 3 males). Maxilla was involved more 80% than mandible. 6:2 Rt side of maxilla was involved more than left side. All the cases presented with facial deformity and extra oral swelling. In all the cases multiple teeth adjacent to lesion failed to erupt. All the 8 cases proved to be intra osseous follicular type of AOT on histopathology. No recurrence was seen after surgical excision, follow up was done for two years.

DISCUSSION

Adenomatoid odontogenic tumor (AOT) is a relatively uncommon distinct odontogenic neoplasm that was first described by Steensland in 1905.[11] However, variety of terms has been used to describe this tumor.

Unal et al[12] produced a list containing all nomenclatures for AOT reported in the literatures. Many different names like adeno ameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma adamantinum or teratomatous odontoma have been used in the

past to define the lesion. Philipsen and Birn[9] in 1969 introduced the now generally accepted and very popularly used nomenclature of AOT.

The debate of whether AOT is an anomalous developmental hamartomatous growth or a true benign tumor has not been settled yet. As AOT has limited size in most cases and has lack of recurrence, it gives predilection hamartoma.[13] Assumption as a tumor[14] is because of the belief that the limited size of most cases stems from the fact that most are detected early after a routine radiograph and removed before the slow growing tumor reaches a clinically noticeable size. Many reported chronic cases had resulted in facial asymmetry and distortion that rival many ameloblastoma.[15,16] Presesnt study reported marked facial deformity in almost all cases.

From the early 1990s onwards 65 single cases of AOT (excluding case series of more than 10 cases) have been published. Age distribution shows that more than two third were diagnosed in the second decade of life and more than half of the cases occurred within the teens (13-19 years of age).

AOT is commonly encountered in young patients especially in second decade of life, and is uncommon above the age of 30 years[16] which is similar to present study. Females were more affected than males with female:male ratio 1.9.[1,9,16] Higher

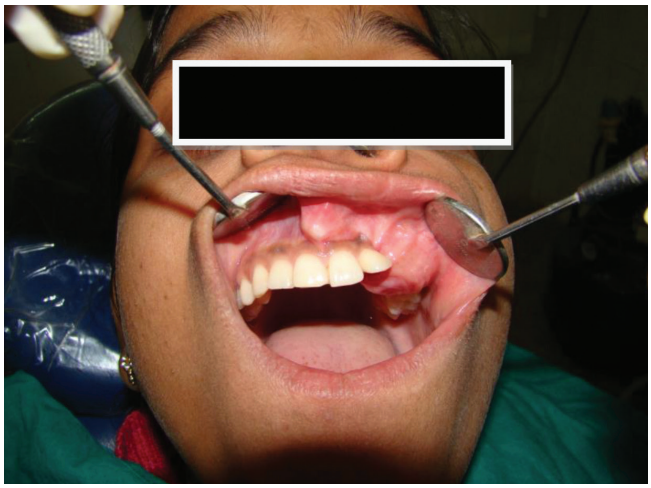

Figure-1: Intra oral photo showing missing canine, first and 2nd premolars

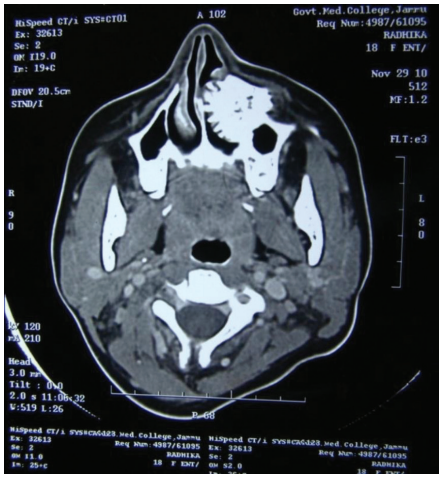

Figure-2: CT Scan showing extent of lesion

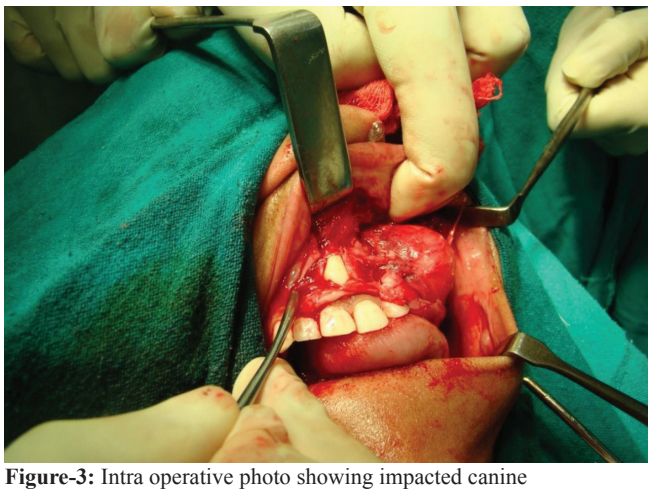

Figure-3: Intra operative photo showing impacted canine

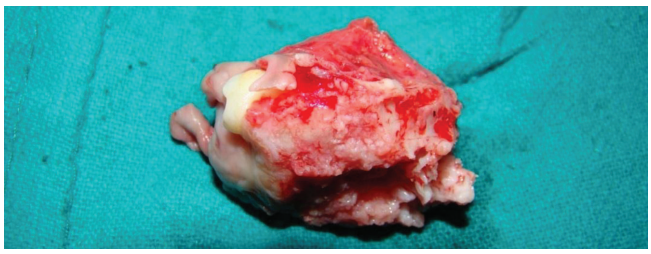

Figure-4: tumor mass with impacted premolars

ratio was found in Asian population, highest incidence being observed in Sri lanka (3:2;1)[13] and Japan (3:1).[3] The present study also reported female involvement more than males.

There are three clinic pathologic variants of AOT, namely intra osseous follicular, intra osseous extra follicular and peripheral, all with identical histology. The follicular type is a central intra osseous lesion associated with an impacted tooth, while extra follicular intra osseous AOT intra osseous AOT has no relation with a UN erupted tooth. In spite of this, it is often located between, above or superimposed upon the roots of adjacent erupted teeth. The peripheral variant appears as a gingival fibroma or pupils attached to the labial gingival[5]

The follicular and extra follicular variants account for 96% of all AOT cases (of which 71% are follicular). Follicular and extra follicular variants together are more commonly found in the maxilla than in the mandible (2.1:1 ratio). Maxilla is predominant site of occurrence, being almost twice as frequent as to that of mandible 2.1;[17], similar to present study 80% cases were reported in maxilla and all were intra osseous follicular variant.

Clinical features generally focus on complaints regarding a missing tooth. The lesion usually present as asymptomatic swelling, which is slow growing and often associated with a unerupted tooth. However, the rare peripheral variant occurs primarily in the gingival tissue of tooth-bearing areas.[18] Unerupted permanent canine are the teeth most often involved with AOTs. Anterior part of the jaw is more frequently affected than posterior region. Tumor is usually associated with unerupted canine and lateral incisor, resorption of roots is rarely seen.[9]

The radiographic findings of AOT frequently resemble other odontogenic lesions such as dentigerous cysts, calcifying odontogenic cysts, calcifying odontogenic tumors, globule-maxillary cysts, ameloblastoma, odontogenic keratocysts and periapical disease.[19] Whereas the follicular variant shows a well circumscribed unilocular radiolucency associated with the crown and often part the root of an unerupted tooth, the radiolucency of the extra-follicular type is located between, above or superimposed upon the roots of erupted permanent teeth.[20] Displacement of neighboring teeth due to tumor expansion is much more common than root resorption. Some erosion of the adjacent cortical bone may occur in peripheral lesions.[9] Remarkably, all variants of AOT show identical histology.The histological typing of the WHO defined the AOT as a tumor of odontogenic epithelium with duct like structures and with varying degree of inductive changes in the connective tissue. The tumour may be partly cystic and in some cases the solid lesion may be present only as masses in the wall of a large cyst.[21] Moreover, eosinophilic, uncalcified, amorphous material can be found and is called "tumor droplets". Some tumor droplets show a homogenous matrix whereas most tumor droplets reveal electron-dense plaques, most probably represent some form of enamel matrix.[15]

A review of literature depicts a lesion in which conservative management produces a uniformly excellent outcome without recurrence.[22,23] Since adenomatoid odontogenic tumor is benign, presents with non aggressive biologic behavior, progressive growth, small frequency of recurrence, absence of invasion and frequent presence of capsule the treatment should consist of enucleation and curettage.[24] Conservative surgical enucleation is the treatment modality of choice. Recurrence of AOT is very rare. Only three cases in Japanese patients are reported in which the occurrence of this tumor occurred therefore, the prognosis is excellent when completely removed in toto.[24] All variants of AOT are well encapsulated and show an identical benign behavior. Surgical enucleation/ excision or curettage is the treatment of choice with only rare recurrence.[9] In the present study all the cases were treated by enucleation / excision and removel of impacted teeth. Follow up was done for 2 years almost, no recurrence was reported

CONCLUSION

By definition a neoplasm has un limited growth potential, The larger size of these lesion supports the classification of AOT as a benign neoplasm and not a hamartoma. It is an un common odontogenic lesion seen in adolescent females. It should be managed at an early stage and excised in toto to prevent damage to the adjacent teeth. Recurrence is very rare. There is important need to report similar cases and such other cases to increase the literature bank and contribution in the maxillofacial specialty.

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