CASE REPORT

A Rare Case of Extrafollicular Adenomatoid Odontogenic Tumour in the Posterior Region of the Mandible: Misdiagnosed as Residual Cyst

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ABSTRACT

Adenomatoid odontogenic tumor is a relatively uncommon distinct odontogenic neoplasm. It is an uncommon tumor of odontogenic origin with varying number of ductlike structures and inductive changes in the stroma. It is a benign and slow growing epithelial tumor and represents 3% of all odontogenic tumors. Its occurrence is more common in anterior region of the maxilla than mandible. Most of the adenomatoid odontogenic tumors occur intra-osseously but few peripheral variant have been reported which are attached to the gingival structures. The intra-osseous Adenomatoid odontogenic tumor may be related to unerrupted tooth (follicular varient) or may not (extrafollicular varient) be related to unerrupted tooth. This paper is to present a rare case of an extrafollicular Adenomatoid odontogenic tumor occurring in the body of the mandible in a male patient which is distinct and secondly it was clinically and radiographically diagnosed as residual cyst. The diagnosis of Adenomatoid odontogenic tumor was confirmed by Histopathological investigation.

Key Words: Adenomatoid Odontogenic Tumour (AOT), Intraosseous, Extrafollicular.

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Introduction

Steensland in 1905 was the first to described Adenomatoid odontogenic tumor (AOT).[1] However initially various terms like adenoameloblastoma, adamantinoma, ameloblastic adenomatoid tumor, ameloblastic adenomatoid tumor or epithelioma adamantinum had been used to describe it but then in 1969 Philipsen and Birn proposed the term AOT and emphasised that it is not a variant of ameloblastoma but a separate entity.[2-5] In 1971 WHO included the term AOT in its first classification of odontogenic tumors1. Although it is classified as benign neoplasm some of the investigators believe it to be hamartomatous malformation due to small size and lack of recurrence.[6] They usually occur intra-osseously, but very few cases of gingival AOT have been reported. Intra-osseously they might be attached to the neck of unerrupted teeth in a true follicular manner[5] or they might have no association with impacted crown as seen in extrafollicular lesions.

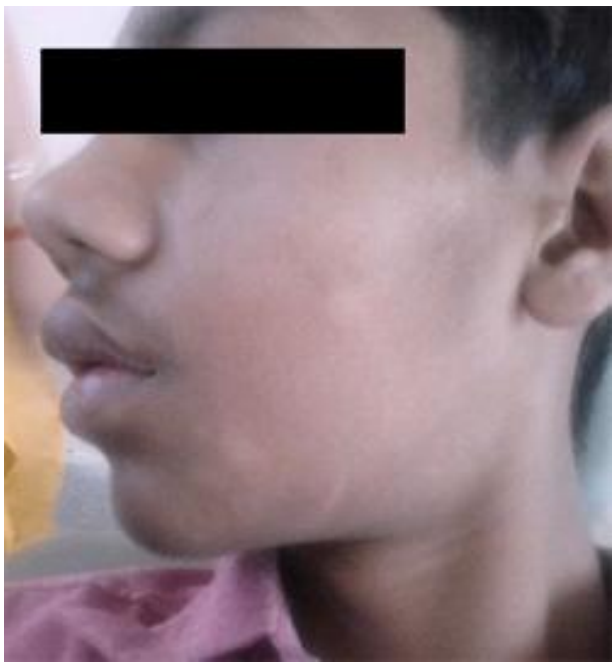

Fig. 1: Extra-Oral Swelling.

Here, we present an uncommon case of extrafollicular variant of AOT in male patient, occurring at unusual site posterior region of the mandible, which was clinically and radiographically diagnosed as residual cyst but histopathologically it was confirmed as AOT.

Case History:

An 18 Year old boy visited the Department of Oral Medicine and Radiology, with a chief complaint of swelling on his left side of the lower jaw since one year, first molar of the same side was extracted at private clinic six months back but the swelling remained. The swelling was slow growing, it gradually increased in size and led to disfigurement of the face. The systemic examination of the patient revealed neither pathological nor systemic disorder. Extra orally (Fig.1) facial asymmetry was seen. A solitary diffused swelling measuring 3.5x2.5 cm in size on the left side of the face was palpable. The swelling extended superio-inferiorly from the corner of the mouth to the inferior border of the mandible and anterio-posteriorly it extended 1 cm. from the left corner of the mouth to the 1cm in front of posterior border of the ramus of the mandible. On palpation the swelling was nontender, hard in consistency and fixed to the underlying bone. Intra oral examination revealed a solitary swelling in the left mandibular ramus and retromolar area with obliteration of the vestibule. Superio-inferiorly it extended from the gingival margin of the lower teeth into the vestibule, anterio-posteriorly it extended from distal margin of lower left first molar up to the retromolar region. The colour of the overlying mucosa was normal. The margins were ill-defined and diffused, the consistency was variable ranging from rubbery to hard in nature. The second molar tooth showed grade-II mobility and the third molar was impacted.

An Orthopantomograph (Fig.2) revealed an unilocular,

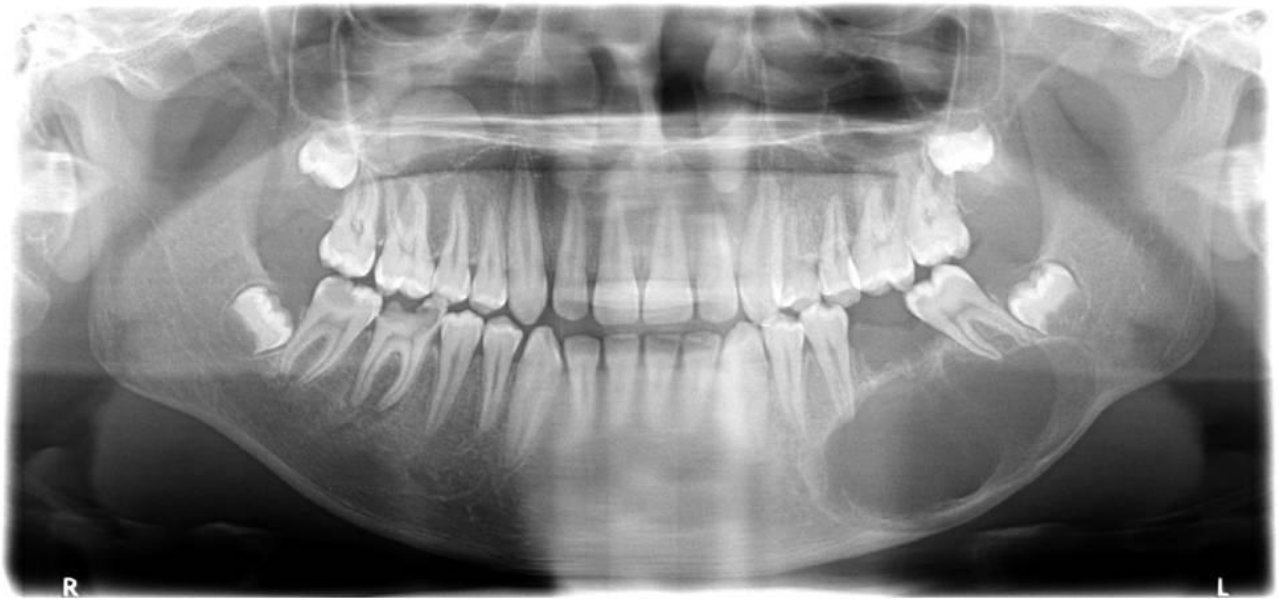

Fig. 2: An Orthopantomograph.

oval radiolucency of 4x5cm in the left body of the mandible region with well defined sclerotic borders with missing first molar tooth, the roots of the second molar tooth drifted distally with no signs of root resorption and impacted third molar (not in relation with the lesion) .

An excisional biopsy was done. Specimen was sent for histopathology. Histopathology revealed densely

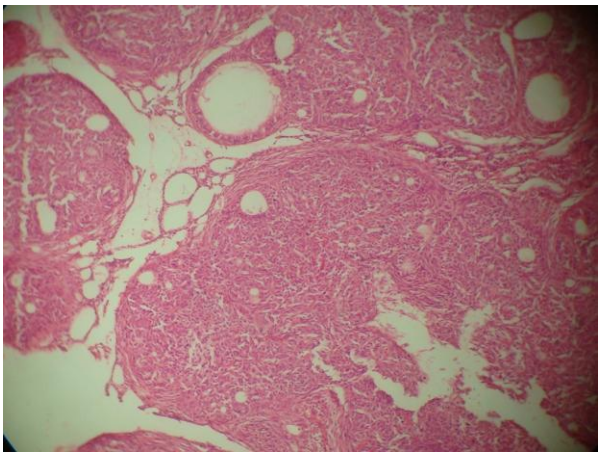

Fig. 3: H&E Stained section. Scanner view 4x

arranged cuboidal to spindle shaped odontogenic epithelial cells in variable pattern like ductal,rosettes and convoluted (Fig-3&4). Scattered small globular to irregular calcifications are seen in the proliferative tissue, features suggestive of Adenomatoid odontogenic tumour (AOT). Thus the final diagnosis was given as extrafollicular AOT.

Discussion:

Adenomatoid odontogenic tumor (AOT) is an odontogenic epithelium tumor with varying number of ductlike structures and lumina of varying sizes (frequently these lumens are lined by hyaline rings), they also show varying degrees of inductive change in the stroma.[1,2,4,7] It is an uncommon, benign, asymptomatic, slow growing tumor with marked predilection for occurrence in the anterior region of the maxilla. It represents 3% of all odontogenic tumors.[2,4,7] It is more commonly found in young patients with two thirds of all cases occurring between age group 10 to 19 years of age.[2,4] It is more common females than males with incidence of almost 2:1.[7-9] Here we report an unusual case of male patient with AOT in the body of the mandible, which was slow growing and was diagnosed clinically as residual cyst. It was initially clinically diagnosed as residual cyst because of the extraction of first molar six months back.

AOT are usually asymptomatic, sometimes may cause cortical expansion and displacement of the adjacent teeth,[4,10,11] as in the case reported here. The slow growing nature of the lesion may cause the patient to tolerate the swelling for years until it produces an obvious deformity.[4,12]

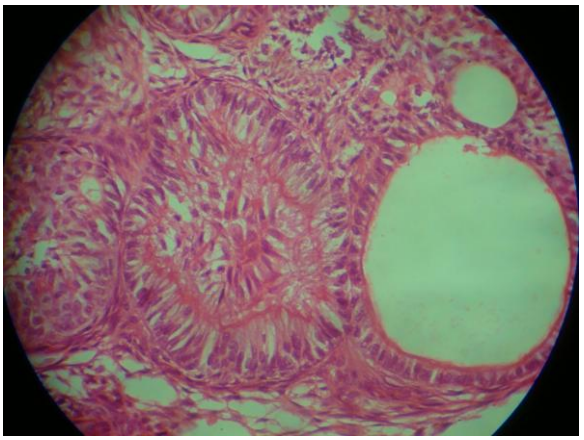

Fig. 4: H&E Stained section showing cuboidal to spindle shaped odontogenic epithelial cells in variable pattern like ductal, rosettes and convoluted. [40x]

Three clinicopathologic variants of the tumor have been described, namely- Intraosseous follicular (73% of all AOT cases), Intraosseous extrafollicular(24%), and (3%)Peripheral[4,11,13] all these variants have identical histological features. The follicular type is the one which is most common with a central intraosseous lesion associated with an impacted tooth. Usually association of the tumor with an impacted tooth is an important feature which makes one think of a diagnosis of adenomatoid odontogenic tumor. Rarely extrafollicular intraosseous AOT which has no relation with unerrupted tooth has been reported. The extrafollicular type is subdivided into E_1 no relation to tooth structure either erupted or unerrupted, E_2 intra radicular adjacent roots diverge apically due to tumor expansion, E_3 superimposed on the root apex, E_4 superimposed on the midroot level[4,11] In the present case there were no impacted teeth associated with the lesion and the impacted 3rd molar was not in relation with the lesion, so the diagnosis of extrafollicular was given.

Although many case have been reported in the literature where AOT is related to other odontogenic

tumors and cysts like ameloblastomas, dentigerous cyst and periapical lesions.[5,6,10] The exert relation of AOT in this case with any other odontogenic or non-odontogenic lesion is not clear because patient was not able to give a details of extracted first molar. His chief complaint has always been facial deformity in the area.

The origin of the AOT is controversial. The histogenesis of AOT is unknown therefore some investigators consider it as hamartoma. But because of its predilection for tooth-bearing bone, it is thought to arise from odontogenic epithelium.[11,13,14] Some investigators believe the possible source of origin of the AOT in the permanent molar region could be due to epithelial remnants from dental lamina and the accessional dental lamina (sometimes together called the parent dental lamina).[11-13]

All variants of AOT show remarkable identical histology.[1] The histological typing of the WHO defined the AOT "as a tumor of odontogenic epithelium with duct-like structures and with varying degrees of inductive change in the connective tissue. The tumor is well encapsulated and shows an identical benign behaviour".[15-19]

Radiographically, it usually appears as well-demarked unilocular radiolucent area located pericoronally or juxtacoronally but sometimes it may extend apically beyond cement-enamel junction specially towards one side of the root. Many a time's small radiopaque foci are seen within the radiolucent lesions, although root resorption is rare.[20-22] Usual unilocular radiolucent area exhibits a well defined corticated or sclerotic border[7] but in case of multiocular radiolucency the boarders are scalloped.[23-26] The patient described in this report has no root resorption, but there is displacement of the adjacent teeth and also the tumor was not associated with an impacted tooth.

The most common treatment modality for the tumour is enucleation and currettage.[11,27] Conservative surgical excision is usually done because the tumor is not locally invasive, it is well encapsulated, and is separated easily from the bone.[1-3,5,8,28] It lacks recurrence with low rate of 0.2%.[2-4,29-30]

Conclusion:

We present a rare case of extra follicular variant of AOT in the posterior left part of the body of the mandible in a male patient with a history of extracted first left mandibular molar six months back. Based on clinical and radiographic examination initial diagnosis of residual cyst was given. The histopathological investigation confirmed the diagnosis of AOT. So the diagnosis of adenomatoid odontogenic tumor is usually confirmed by histopathological finding, as clinical and radiographical features may sometime be misleading. AOT must be considered in the differential diagnosis of corticated radiolucencies with or without small radiopaque foci, especially in teenagers and young adults, even in the absence of an impacted tooth.

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