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An unusual site of Adenomatoid Odontogenic Tumor: A rare case report

Sapna Panjwani*, Anjana Bagewadi, Vaishali Keluskar*****

*Post Graduate Student **MDS ,Professor, ***Professor and Head, Department of Oral Medicine and Radiology , KLES VK Institute Of Dental Sciences, Belgaum, Karnataka.
Contact: dr.sapna28@gmail.com

Abstract

The adenomatoid odontogenic tumor (AOT) is a rare odontogenic tumor constituting only 3 % of all the odontogenic tumors and often misdiagnosed as an odontogenic cyst. Though odontogenic in origin but the presence of so called "duct like structures" is a unique microscopic feature which gives the lesion a glandular, ie. Adenomatoid appearance.

Here we are presenting a rare case report of an extrafollicular adenomatoid odontogenic tumor in the mandible w.r.t 31, 32, 33 and 34, which is an unusual site for the same. However, the diagnosis of an adenomatoid odontogenic tumor should be considered when the clinician is presented with a corticated radiolucency in the anterior lower jaw, especially in teens and young adults.

Keywords: Benign, Odontogenic Tumor, Unilocular Radiolucency, Extrafollicular, Adamantinoma.

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Introduction:

Adenomatoid odontogenic tumor (AOT) is a benign (hamartomatous), non-invasive lesion with slow but progressive growth that accounts for 2.2–13% of all odontogenic tumors. AOT usually affects young patients, mostly during their second decade of life. There is a tendency to affect females (male: female ratio 1:2) and to occur in the anterior maxillary region.[1, 2, 3, 4]. It was first described by Driebladt in 1907 as a pseudoadenoma adamantinoma.[5]

There are three variants of AOT: follicular, extrafollicular, and peripheral. The follicular type is a central intrabony lesion associated with an unerupted tooth, which accounts for about 70% of all cases. The extrafollicular type is also an intra-osseous lesion, but unrelated to an unerupted tooth, and represents 25% of all AOTs. The peripheral type is a rare form that arises in the gingival tissue, and only 18 well-documented cases were reported.[2, 4] All three variants have the same histological aspect and clinical behavior[6].

This case paper aims to describe an unusual case of AOT, originating in the mandibular anterior region of an 18-year-old female.

Case Report:

An 18 year old female patient reported to the Department of Oral Medicine and Radiology, with a chief complaint of growth in left lower jaw region since 1 year. History revealed the presence of similar growth in the same region 2 years back which was surgically excised but had recurred. Growth had progressively increased to attain the present size with no associated pain, discharge and numbness revealed.

Intraoral examination revealed a solitary growth measuring about 2X2 cms situated on the attached gingiva extending from distal aspect of 32 to mesial aspect of 34 (Figure 1).

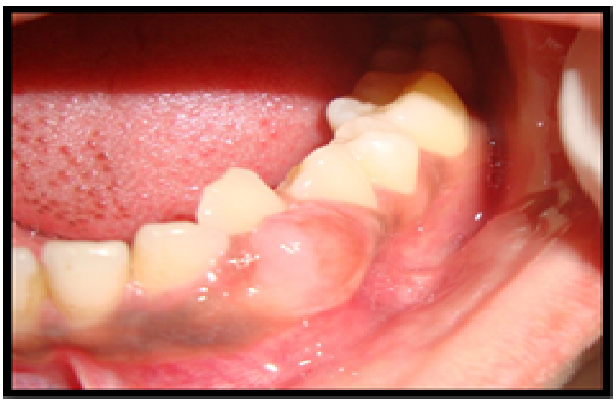

Fig 1: Intraoral gingival growth w.r.t 33, 34.

Mucosa over the growth was pale and fibrosed. On palpation, the growth was firm in consistency and non-tender. Another diffuse swelling was present obliterating the buccal and lingual vestibule measuring about 2X2 cms in size extending from 31 – 34 region antero-posteriorly and from base of the growth to the depth of vestibule supero-inferiorly (Figure 2). The overlying mucosa was intact with no ulceration and sinus discharge.

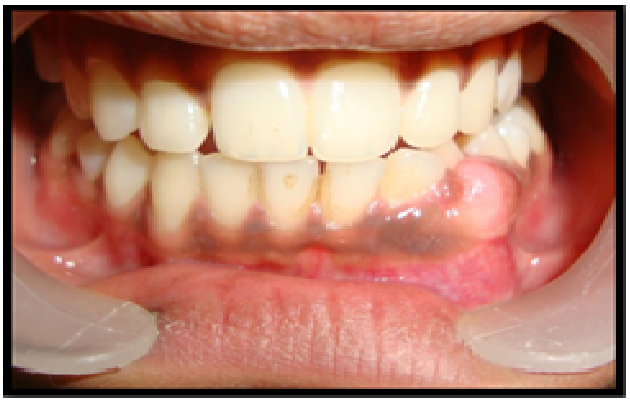

Fig 2: Intraoral swelling w.r.t 31, 32, 33, 34 region

On palpation, swelling was firm in consistency and showed no evidence of discharge on digital pressure. It was non-tender with no pulsations felt. Egg shell crackling was evident on palpation. The associated teeth i.e. 32, 33, 34 and 35 were found to be grade I mobile and nonvital on cold and electric pilp vitality testing.

The patient was subjected for radiographic investigations: Intraoral periapical (IOPA) radiograph w.r.t 31, 32, 33 and 34(Figure 3); mandibular cross-sectional occlusal radiograph (Figure 4) and orthopantomograph (Figure 5).

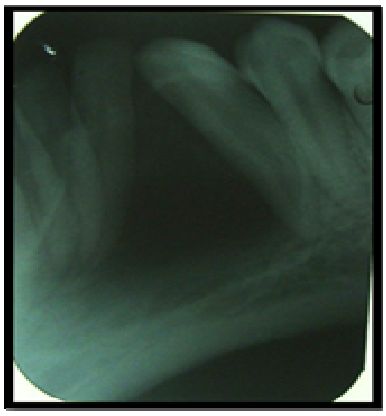

Fig 3: IOPA radiograph showing displacement with 32 and 33 and a well defined radiolucency

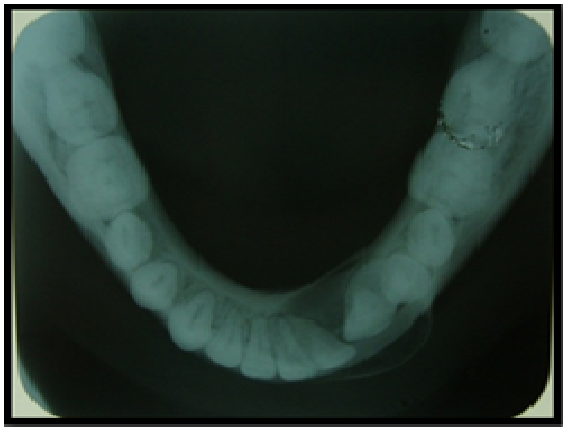

Fig 4: Mandibular occlusal cross-sectional view showing bicortical expansion.

The radiographs revealed a well-defined periapical radiolucency measuring about 2 X 1.5cms with sclerotic border extending from distal aspect of 31 to mesial aspect of 34. IOPA radiograph revealed displacement with 32 and 33 and occlusal radiograph revealed expansion with buccal and lingual cortical plates.

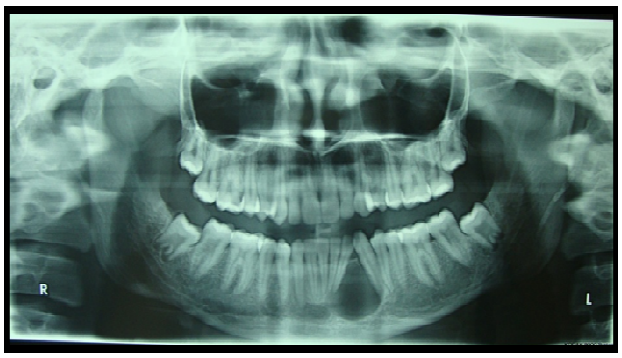

Fig 5: Panoramic radiograph revealing large unilocular periapical radiolucency surrounded by a sclerotic border extending from distal aspect of 31 to mesial aspect of 34. Displacement with 32 and 33 is evident.

Following this the patient was subjected for fine needle aspiration which yielded a straw coloured fluid. Microscopic examination of the same revealed histiocytes, mast cells; polymorph macrophages and few foam cells.

Based on the clinical features and history, provisional diagnosis of adenomatoid odontogenic tumor and traumatic fibroma were given. Various differential diagnosis such as periapical cyst, traumatic bone cyst, aneurysmal bone cyst, ameloblastoma were put forth.

Root canal treatment was done w.r.t 31, 32, 33 and 34. Surgical enucleation of the tumor and the growth were carried out under local anaesthesia and the samples were sent for histopathologic examination. The tumor cavity was packed with 3 large pieces of Gelfoam and the mucoperiosteal flap was replaced in contact with the packing. Healing was uneventful, with no evidence of recurrence after surgery.

Histopathological report (Fig: 5 & Fig: 6) revealed a cystic cavity lined by stratified squamous epithelium with multinodular proliferation of spindle, columnar and cuboidal cells arranged in form of sheets, strands and whorled masses.

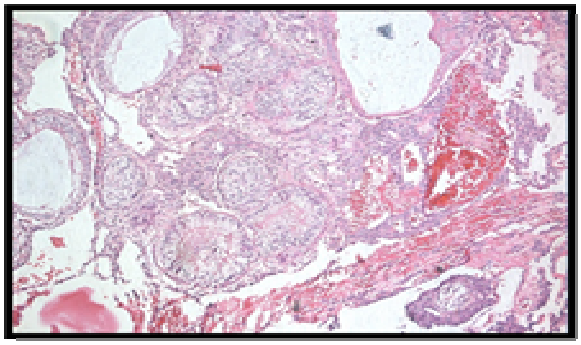

Fig 6:Photomicrograph showing histological features (Hematoxylin-Eosin stain X 4x

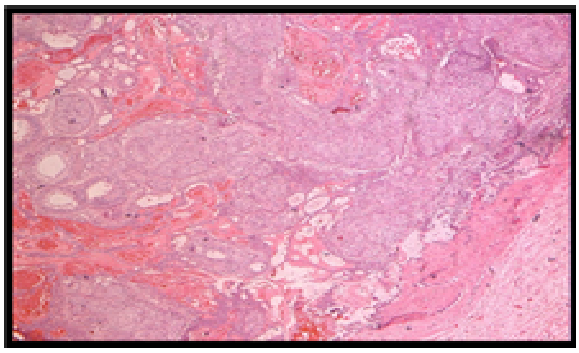

Fig 7: Photomicrograph showing histological features (Hematoxylin-Eosin stain X 10x)

Epithelial cells revealed rosette like structure about a central space containing eosinophilic substance. The characteristic duct like structure with lumen lined by single layer of cuboidal and columnar cells with scattered eosinophilic substance were present, suggestive of adenomatoid odontogenic tumor with respect to anterior portion of left side of mandible.

Discussion:

The AOT comprises approximately 3% of all odontogenic tumors, ranking behind odontoma, periapical cemental dysplasia, myxoma and ameloblastoma[6]. The origin of AOT is controversial. However, evidence also exists that the tumor could be derived from epithelial remnants of the dental lamina complex system[7, 8, 9, 10]. The lesion then presents radiographically as a residual, developmental, lateral periodontal or radicular cyst, depending on the location of the epithelial rest cells.

The case report illustrates characteristic clinical and radiographic features of the extrafollicular variant of AOT at an unusual site (mandible). AOT is prevalent in the second decade of life, female sex being affected twice as often as compared to male sex. The common site of involvement for the follicular variety is maxillary canine region and extrafollicular variant is maxillary incisor or canine region[11]. A distinct radio-opaque border of the unilocular radiolucency is a characteristic radiographic manifestation of AOT. It is usually associated with the displacement of the teeth which was evident in our case. However, the presence of rare feature in our case was egg shell crackling mimicking to be a unicystic ameloblastoma[12]. Root resorption is seldom reported, however it was not present in our case. Conservative surgical enucleation or curettage is the treatment of choice with only rare recurrence (0.2%). The patient we described had no recurrence and a regular follow-up was done after local excision.

Conclusion:

It should be emphasized that extrafollicular variant of AOT is very rare. Only careful diagnosis and adequate interpretation of clinical and radiographic findings may be helpful in arriving at a correct diagnosis. The final diagnosis of an AOT was arrived by histologic examination of the excised tissue specimen.

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