Case Report

Adenomatoid Odontogenic Tumor involving Maxillary Sinus -A Rare Case Report

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Abstract: The Adenomatoid Odontogenic Tumor is a rare, slow growing, benign, odontogenic epithelial tumor with characteristic clinical and histological features; which usually arise in the second or third decade. It is a tumor composed of odontogenic epithelium in a variety of histoarchitectural patterns which are embedded in a mature connective tissue stroma. This case report describes a case of 26 year old female with swelling and occasional mild pain in the upper right canine region. Intraoral examination showed that the labial vestibule was obliterated by expansion of the buccal cortical plate from the upper right lateral incisor to the first molar region. The maxillary right lateral incisor was displaced labially, deciduous canine was retained and permanent canine was missing.

Key words: Adenomatoid Odontogenic Tumor, Histoarchitectural, Obliterated, Retained.

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Introduction

Adenomatoid odontogenic tumor was first described by Dreibladt, in 1907, as a pseudo adenoameloblastoma. In 1948 Stafne considered it a distinct entity, but it was classified by others as a variant of ameloblastoma.[1] Philipsen and Birn proposed the name adenomatoid odontogenic tumour in 1969 and suggested that it not be regarded as a variant of ameloblastoma because of its different behavior. This term was adopted by the World Health Organization (WHO) classification in 1971.[1,2] However, in 2005 the histologic typing of WHO defined AOT as a tumor composed of odontogenic epithelium presenting a variety of histoarchitectural paterns, embedded in mature connective tissue stroma, and

characterized by slow but progressive growth.[3]

The Adenomatoid Odontogenic Tumor (AOT) has been described as an uncommon benign odontogenic tumor with a relative frequency of 2.2 – 7.1%.[4] It is a rare slow growing, benign, odontogenic epithelial tumor with characteristic clinical and histological features which usually arise in the second or third decade. Most AOT occur intraosseously. They surround the crowns and are attached to the necks of unerupted teeth in a true follicular relationship. Some however have no association with unerupted teeth and a few arise on the gingiva in an extraosseous location.[5] The tumor affects females more than males in almost a two to one ratio. The maxilla is involved nearly twice as frequently as the mandible of which the canine region is the most commonly involved region.[6,7] We, hereby report a case of AOT arising from maxillary canine region.

Case Report

A 26-year-old female presented to the Department of Oral and Maxillofacial Surgery, with swelling and occasional mild pain in the upper right canine region. Intraoral examination showed that the labial vestibule was obliterated by expansion of the buccal cortical plate from the upper right lateral incisor to the first molar region. The maxillary right lateral incisor was displaced labially, deciduous canine was retained and permanent canine was missing (Figure 1).

Figure 1: Intraoral examination showing the lesion.

Panoramic radiograph revealed a large, well-circumscribed radiolucency extending throughout the anterior maxilla from the right lateral incisor to the right first molar region involving maxillary sinus with impacted canine (Figure 2). The lesion produced an expansion of tissue and extended into the alveolar processes, disrupting the usual orientation of the anterior teeth and first premolar tooth.

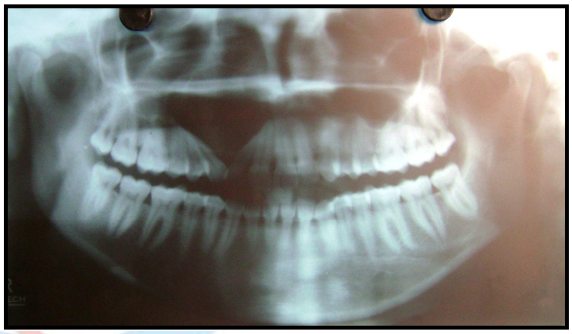

Figure 2: Panoramic radiograph revealed a large, well-circumscribed radiolucency.

On palpation the swelling was soft in consistency without any discharge of fluid. Following this the patient was subjected for FNAC which yielded with straw colored fluid but after aspiration lesion reduced its softness. On the basis of the clinical and radiographic findings, the differential diagnosis was dentigerous cyst, adenomatoid odontogenic tumour, calcifying odontogenic cyst and calcifying epithelial odontogenic tumour. The patient underwent surgery with local anesthesia. A crevicular incison was given; mucoperiosteal flap from the right molar region was reflected to expose the labial aspect of the tumour. The

labial cortex was very thin and had several areas of complete resorption (Figure 3).

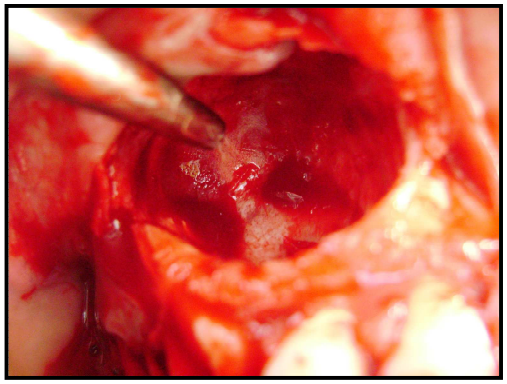

Figure 3: Surgical procedure to remove the lesion.

The tumor was removed and it was tennis ball like rounded lesion (Figure 4).

Figure 4: Gross appearance of tumor appearing like a tennis ball.

The areas between the roots of the involved teeth were curetted well. Impacted canine was not involved in lesion; it was away from lesion and was removed after removal of tumor. The cavity was packed with iodine gauze and the flap was sutured in place. Patient was recalled after one week. Histopathological examination revealed sheets of polygonal and spindle shaped cells proliferating in multiple duct like patterns throughout the fibrous connective tissue stroma. The ductal lumina were filled in some areas with eosinophilic material, although some lumens were empty. The duct like structure exhibits a central space, which is bordered on the periphery by single layer of columnar cells resembling ameloblasts. It was diagnosed as Adenomatoid odontogenic tumor (Figure 5).

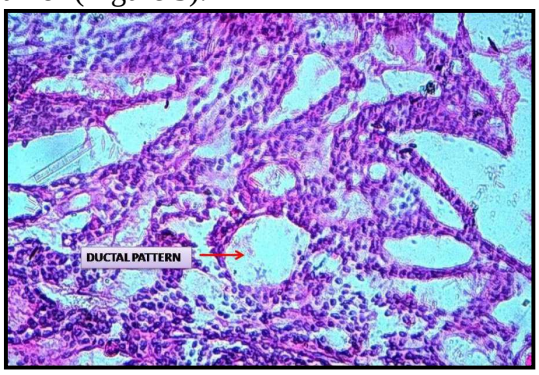

Figure 5: Histological appearance showing duct like pattern.

Healing was uneventful, and there was no evidence of recurrence 1 year after the surgery.

Discussion

The AOT is an uncommon cause of jaw swelling.[8] There is a slightly female over male incidence, an almost 2:1,[9] and appears most often in the second and third decade of life.[5] The sex and the age of the patient we described in this report was consisted with the literature. The lesions are typically asymptomatic, but may cause cortical expansion and displacement of the adjacent teeth,[1] as in the case reported here. AOT involves both the bone and soft tissue in anatomic configuration.[10,11] Our case followed the biological trend of the common intraosseous location in the maxillary canine region. Cases have been reported in the mandible, molar areas, in maxillary sinus and along with embedded primary teeth.[12] The origin of the AOT is controversial. Because of its predilection for tooth-bearing bone, it is thought to arise from odontogenic epithelium.[13,14] The histological typing of the WHO defined the AOT as a tumor of

odontogenic epithelium with duct like structures[15] as in our case and with varying degrees of inductive changes in the connective tissue. The tumor may be partly cystic, and in some cases the solid lesion may be present only as masses in the wall of a large cyst. 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,[16] most probably represent some form of enamel matrix.[17] Conservative surgical enucleation is the treatment modality of choice. Recurrence of AOT is exceptionally rare, the prognosis is excellent when completely removed in toto.[16]

Conclusion

Careful diagnosis and adequate interpretation of clinical and radiographic findings are helpful in arriving at correct diagnosis. But usually the final diagnosis is arrived by histological examination of excised tissue specimen as in our case.

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