Case Report

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A Case of Two-Third Tumour In Maxilla: A Case Report

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ABSTRACT

Background: Adenomatoid Odontogenic Tumor (AOT) represents 2% to 7% of all odontogenic tumors. It is an uncommon benign tumor of odontogenic origin composed of odontogenic epithelium in a variety of histopathological patterns. These lesions are usually solid but are occasionally cystic. We present a rare case of AOT arising from an odontogenic cyst around the impacted canine in an 18-year-old female. We believe that this case is an odontogenic cyst with neoplastic development, containing both epithelial and mesenchymal components. It is slow-growing, benign in nature that appears in the anterior portion of the jaws and more frequently in the anterior maxilla and more in females and associated with the impacted canine. The treatment of choice was surgical management of the tumor, and the final diagnosis is purely based on the histological examination. The present report describes a rare case of AOT associated with an odontogenic cyst occurring in the anterior region of the upper jaw with an impacted left canine, highlighting the importance of the histopathological examination in the cystic lesions of the jawbones.

Keywords: Maxillary, tumor, Adenomatoid Odontogenic Tumor.

INTRODUCTION

AOT is an uncommon benign tumor of odontogenic origin composed of odontogenic epithelium in a variety of histopathological patterns. These lesions are usually solid but are occasionally cystic. It has been reported that some odontogenic cysts that occur in association with odontogenic tumors of epithelial lining from the cyst may transform into odontogenic neoplasm like ameloblastoma or AOT. Because neoplastic and hamartomata's lesions can occur at any stage of odontogenesis, odontogenic tumors with combined features of epithelial and mesenchymal components may arise within the odontogenic cyst. In this case, the odontogenic cyst was found in the same lesion.

CASE REPORT

An 18-year-old female patient reported to the Department of Oral and Maxillofacial Surgery with a chief complaint of pain in the left front tooth region of the upper jaw for two months. The associated symptoms included discomfort during the chewing of food. The patient was asymptomatic before two months, and the swelling was slow-growing and increased to the present size. Dull, intermittent pain and aggravated on having cold foods. Intraoral examination revealed a well-defined swelling of 3 × 3 cm size in the buccal vestibule extending from the lateral incisor to the second premolar and retained 63(canine) and missing permanent canine. On palpation, the swelling was firm, tender and showed egg-shell crackling, showed no evidence of discharge on digital pressure. The overlying mucosa was blanching and non-ulcerated (fig.1 and 2). Based on the history and clinical features, a provisional diagnosis of a dentigerous cyst was made. Differential diagnosis as a calcifying epithelial odontogenic cyst, adenomatoid odontogenic tumour.

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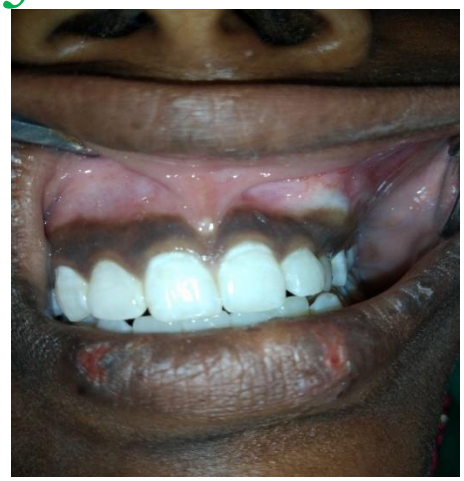

Fig 1: Clinical photograph showing swelling.

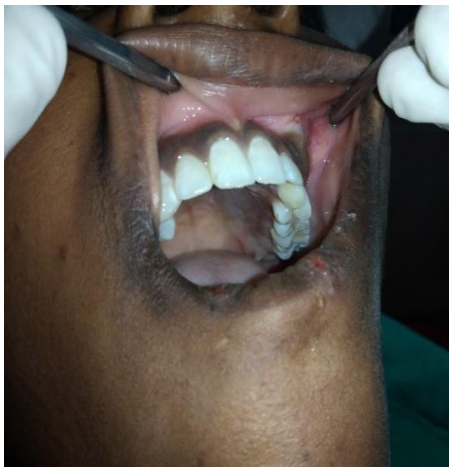

Fig 2: Clinical photograph showing retained 63.

Complete blood investigations were performed, and fine-needle aspiration cytology was done. During FNAC, straw colour fluid was obtained. The cytological report gave a differential diagnosis as an Odontogenic cyst. On Cone-beam computed tomography (CBCT) examination (fig.3): well-defined unilocular radiolucency associated with the crown of unerupted tooth 23, which is extended from distal aspect of 22 to mesial aspect of 25, is seen in coronal section (fig.4). unilocular radiolucency with buccal cortical plate expansion in axial section (fig5). Cystic wall expansion below and above the CEJ of 23, root resorption of retained 63 and tooth displacement of 24 and well radio-opacities has seen within the cystic wall in the sagittal section (Fig.6). The above CBCT findings, it reveals as an adenomatoid odontogenic tumour

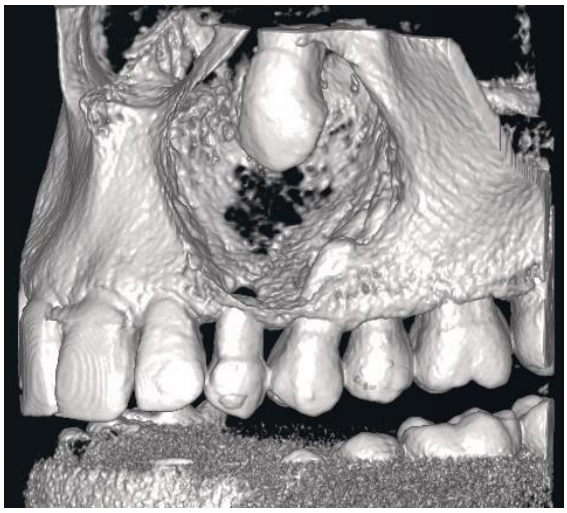

Fig 3: CBCT.

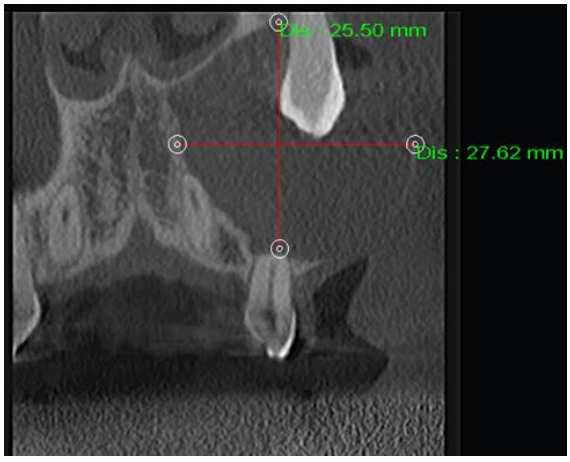

Fig 4: Coronal section.

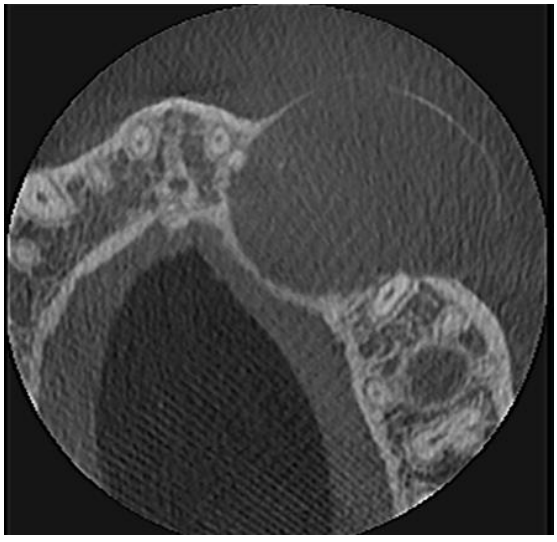

Fig 5: Axial section.

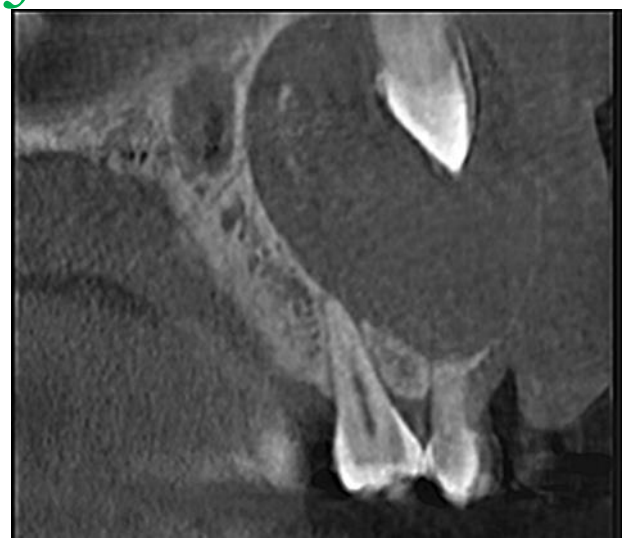

Fig 6: Sagittal section

Surgical procedure

The patient was operated on under 2% local anaesthesia. The crevicular incision was given from 22 to 24 region, and a buccal mucoperiosteal flap was raised. Retained deciduous 63 was removed. The expansion was noted in the left maxillary region with egg-shell thinning of the buccal cortex. The flap was gently teased off the cortex, and the lesion was visualised. It was firm and well encapsulated on palpation. The lesion was enucleated with the tooth in toto. The residual defect was curetted, debrided and the flap sutured in position (fig.7). The specimen (fig.8) was sent to the histopathological examination.

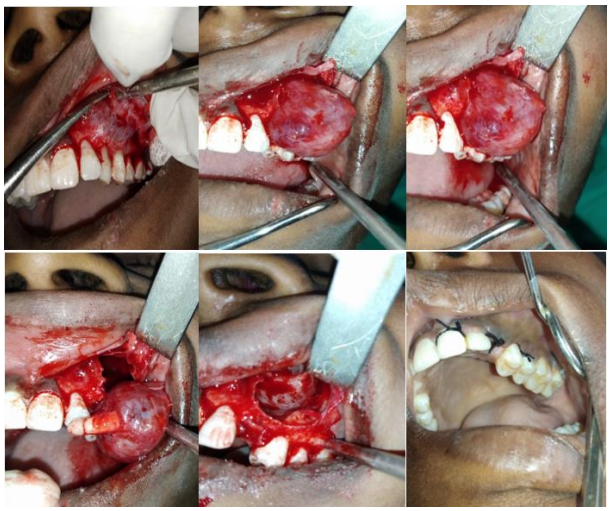

Fig 7: Surgical procedures.

Histological examination (fig.9) reveals 2-3 layers of non-keratinized flattened epithelial lining with multiple nodules of rosette like arrangements and sheets of spindle shaped epithelial cells with hyper chromatic nuclei. Focal areas show duct-like structures with a lining of columnar cells. Connective tissue is fibrovascular with numerous endothelial lined blood capillaries and dystrophic calcifications. These features are suggestive of an adenomatoid odontogenic tumour.

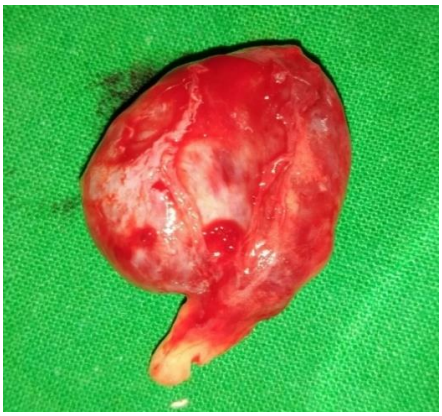

Fig 8: Tumor.

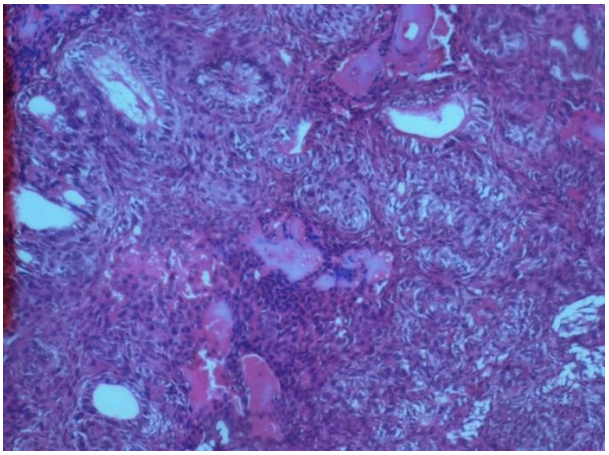

Fig 9: Histopathological section.

DISCUSSION

AOT represents 2% to 7% of all odontogenic tumours, and more than 750 examples have been reported in the literature. Although this lesion was formerly considered a variant of the ameloblastoma and was designated as “adenoameloblastoma,” its clinical features and biologic behaviour indicate that it is a separate entity[6]. AOT, an uncommon benign epithelial lesion of odontogenic origin, was first described by Dreibaldt in 1907 as ‘Pseudoadenoameloblastoma.’ Harbitz, in 1915, reported it as ‘Cystic Adamantoma.’ Stafne, in 1948, considered it a distinct entity, and the term

"Adenomatoid Odontogenic Tumor" was proposed in 1969 by Philipsen and Birn. WHO has described AOT as "A tumour of the odontogenic epithelium with duct-like structures and with varying degrees of inductive change 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[1]

Various terms such as adenoameloblastoma, ameloblastic adenomatoid tumour, adamantinoma, epithelioma adamantium, or trachomatous odontoma were used earlier to describe the lesion [4].

The AOT is also called 'two-thirds tumour' because 2/3rd of adenomatoid tumours occur in the maxilla, 2/3rd occur in young females, two-thirds of the cases are associated with un-erupted teeth, and two-thirds of the affected teeth are canines [2]. It was first described by Dreibaldt in 1907 as pseudo-adenoameloblastoma.

AOT was first recognized as a distinct pathological entity by Stafne in 1948. There are three variants of AOT based on clinical and radiological features as follows:

a. The follicular type has a central lesion associated with an embedded tooth.

b. The extrafollicular type has a central lesion and no connection with the tooth.

c. The peripheral variety[3].

It has been reported that some odontogenic cysts that occur in association with odontogenic tumours of epithelial lining from the cyst may transform into odontogenic neoplasm like ameloblastoma or AOT. Because neoplastic and hamartomatous lesions can occur at any stage of odontogenesis, odontogenic tumours with combined features of epithelial and mesenchymal components may arise within the odontogenic cyst[7].

In this case, AOTand odontogenic cyst was found in the same lesion. Clinical, radiographic, and microscopic findings in the case were consistent with previous descriptions of the lesion in the literature. As previously mentioned, AOTs are usually reliable but are occasionally cystic. Our patient was of the cystic variety as its growth was in the walls of the odontogenic cyst[7].

Radiologically, it should be differentiated from the dentigerous cyst, which most frequently occurs as a pericoronal radiolucency in the jaws. Dentigerous cyst encloses only the coronal portion of the impacted tooth, whereas AOT shows radiolucency, usually surrounding both the coronal and radicular aspects of the involved tooth. But in cases where AOT grows from cyst-like in this case, the radiographs are inconclusive. However, they contain fine calcifications (snowflake), a feature that may be helpful in differentiating an AOT from the dentigerous cyst. The unilocular radiolucency is well demarcated with a smooth cortical border. Most lesions are pericoronal, juxta coronal, and divergence of roots and displacement of teeth often occurs without root resorption cyst may indicate the development of AOT as seen in this case[3].

The origin of the follicular variant can occur before or after cystic expansion (Cystic expansion in the jaw bone refers to the nature of the expansion of the cyst through the buccal and lingual/palatal cortical plates). If it occurred after cystic expansion, then it effectively meant that the origin was from a dentigerous cyst, and several such cases have been reported. If it occurred before cystic expansion, then the tumour tissue would fill the follicular space, and the AOT would present as a solid tumour. In this case, the cystic lining was present, with deeper areas showing tumor islands, suggesting that the tumor could have occurred after cystic expansion[2].

Microscopically AOT is composed of spindle epithelial cells forming nests, cords, or cell masses in a little fibrous stroma. Rosettes can be formed from the epithelial cells around a central space that may be empty or containing eosinophilic material. Duct-like structures can be prominent, sparse, or even absent and constitute a central space, surrounded by cuboidal or columnar cells with peripheral nuclei. A small focus of calcification may be scattered throughout the tumor, being interpreted as the abortive formation of enamel/dentine/cementum[5].

Treatment and prognosis

The treatment of choice was the surgical excision of the tumour. It should be enucleated along with the associated impacted tooth and simple curettage. Conservative treatment is adequate because the tumour is not locally invasive, is well encapsulated, and can be easily separated from the bone. The surgical specimen may be solid or cystic. Its clinical behaviour is more like hamartomas than neoplasms.[6]

CONFLICTS OF INTEREST

The authors declare they have no potential conflict of interests regarding this article.

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