Case Report

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Adenomatoid Odontogenic Tumor: A Case Report

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ABSTRACT

Background: Adenomatoid odontogenic tumour (AOT) is a relatively rare and distinct odontogenic tumour that is exclusively odontogenic epithelium in origin. It comprises 3% of all odontogenic tumors. In the literature, it has been considered as a hamartoma rather than true neoplasm because of its limited size, minimal growth potential and lack of recurrence. This case report describes a clinical course, morphological characteristics of AOT in the right maxilla of a 17-year-old female patient.

Keywords: Adenomatoid odontogenic tumour, hamartoma, neoplasm.

INTRODUCTION

The adenomatoid odontogenic tumor generally considered to be an uncommon tumor, occurs mostly in association with an unerupted maxillary canine. Some investigators consider it as benign neoplasm, while others categorized it as hamartomatous malformation due to its limited size and lack of recurrence of most of the cases. [1] Adenomatoid odontogenic tumors represent 2.2 to 7.1% of all odontogenic tumors [2]. Adenomatoid odontogenic tumors are limited to young patients; two-thirds of all cases are diagnosed in 10-19-year-old patients. This lesion frequently has its onset at the anterior section of upper and lower jaws and is related to non-erupted canines. Female patients are twice over affected when compared with male patients. [3] Regarding its pathogenesis, the lesion originates from odontogenic epithelium (enamel organ or dental lamina remnants) with inductive influence on odontogenic ectomesenchyme and the consequent production of dentinoid material. [3] It often causes expansion of surrounding bone and displacement of adjacent teeth. However, the slow-growing nature of the lesion may cause the patients to tolerate the swelling for years until it produces an obvious deformity.[4]

Radiographically, the tumour usually appears as well-circumscribed, unilocular radiolucency, which may be associated with an unerupted tooth, most often a canine. [5] Conservative surgical excision has been considered as the treatment modality of choice. With the above background, this report deals a case of swelling in a 17-year-old female patient diagnosed histopathologically as Adenomatoid odontogenic tumor.

CASE REPORT

A 17-year-old female patient visited the department of oral medicine with the chief complaint of swelling in the right side of the face for the last year. Swelling is associated with mild pain. There was no history of trauma or pus discharge with swelling. The medical and family history was unremarkable. Extraoral, there was facial asymmetry due to diffuse swelling

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present in the anterior maxillary region. There is the obliteration of nasolabial fold in the right side. The margins are mill defined. On palpation, the swelling was warm, slightly tender and firm in consistency skin over the swelling was pinchable. The swelling was not compressible or reducible.

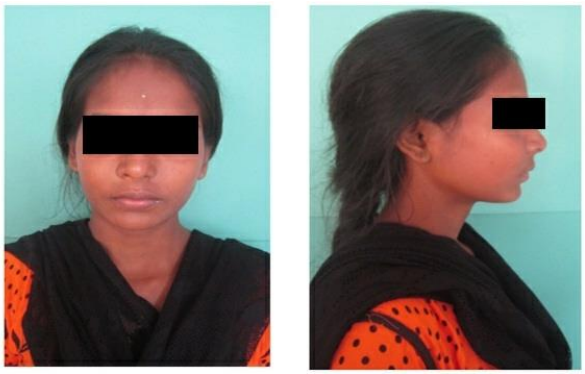


Fig 1: Extraoral photograph showing a diffuse swelling over the right maxillary region.

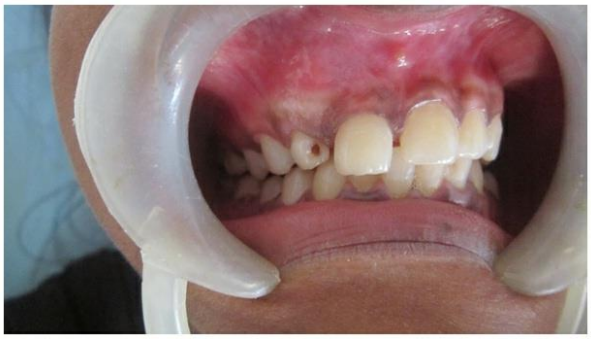


Fig 2: Introoral photograft shows retained deciduous canine (53) missing lateral incisor (12) and canine (13).

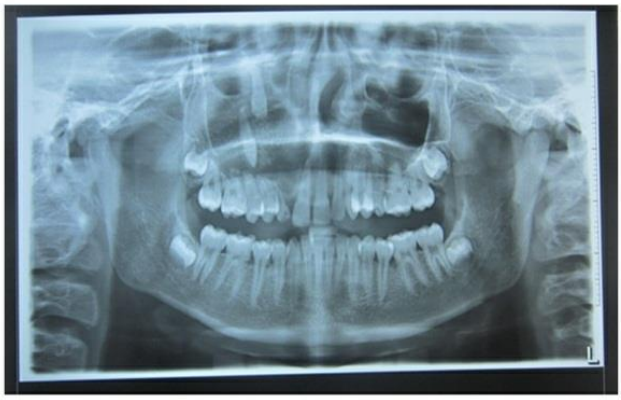
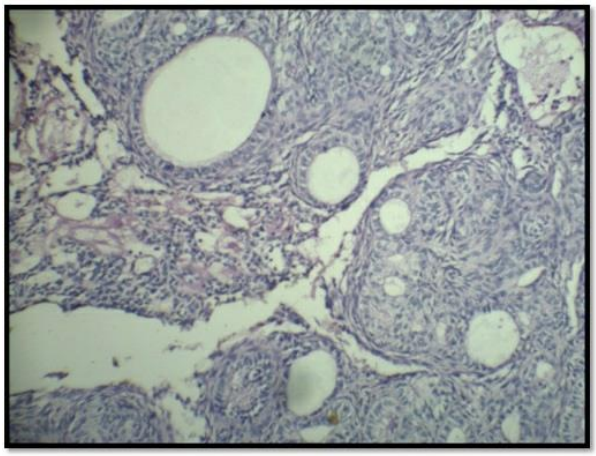


Fig 3: OPG showing well-circumscribed radiolucency with impacted lateral incisor and canine.



4. Photomicrograph showing tumor cells arranged in ductal and rosette pattern.

Fig 4: Photomicrograph showing tumor cells arranged in ductal and rosette pattern.

Intraorally a single diffuse swelling in the alveolar mucosa extends from the mesial aspect of 11 to distal aspect of 14 measuring about 3X2 cm. Retained deciduous right canine 53, missing 12, 13. The swelling extends labially obliterating the labial sulcus. On palpation, all inspection findings were confirmed, the swelling was firm, not fluctuant. The swelling was tender on palpation.

Differential diagnosis:

On the basis of clinical findings, a provisional diagnosis of the adenomatoid odontogenic tumour was given. The following lesions have been considered as differential diagnosis, calcifying odontogenic cyst, Ameloblastoma, calcifying odontogenic tumour, central ossifying fibroma.

Investigations:

Initially, OPG was performed to locate the missing right maxillary lateral incisor and right maxillary canine. OPG reveals a large unilocular radiolucency extends from the mesial aspect of 11 to distal aspect of 14 and impacted 12,13. It was irregular in shape and well-circumscribed with corticated margins. Lateral incisor is present in the floor of the orbit, and canine is located in the maxillary sinus.

Incisional biopsy of the lesion revealed solid sheets of odontogenic epithelial cells composed of duct-like structures lined by cuboidal to columnar cells. The lumen of the duct-like structures shows eosinophilic material. Epithelial cells forming rosette pattern are seen. Interlacing cords and nests

of the odontogenic epithelium was seen. At one focus calcified material seen, connective tissue stroma was scanty.

DISCUSSION

AOT is a relatively uncommon distinct odontogenic neoplasm that was first described by Steensland in 1905.[6] Stafne, in 1948 described it as an «ameloblastic adenomatoid tumor or adeno-ameloblastoma»,since it presented structures which simulated canals or glands, he, therefore, described it as an ameloblastoma variant.[7]. In 1969 Philipsen and Birn proposed the current name: adenomatoid odontogenic tumor (AOT). This name was accepted by the World Health Organization (WHO) in 1971.

The pathogenesis of AOT is unknown. The cell of the origin was suggested as entrapped epithelium in the line of fusion by Stafne (1948); odontogenic epithelium, outer enamel epithelium, inner enamel epithelium, cell rests of Malassez, dental lamina or its remnants.[8,9] Adenomatoid odontogenic tumors present three clinical variants; follicular, extra-follicular and peripheral. The follicular type is characterized by being intra-osseous and associated to a non-erupted tooth. Due to its radiographic characteristics, it is oftentimes confused with dentigerous cysts, since there is the presence of a well-defined radio-lucid area surrounding root and crown of a non-erupted tooth.

The extra-follicular type is characterized by being intra-osseous, and unrelated to a non-erupted tooth. The peripheral type is extra-osseous and appears as an increase in gingival soft tissue volume.[10] AOT is a slow-growing lesion with a predilection for the anterior maxilla associated with impacted canine of young female in a second decade of life. The lesions are usually asymptomatic, but may cause cortical expansion and displacement of adjacent teeth , as in the case reported here.

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.[10,11].

As the tumor is well encapsulated and benign in nature, conservative surgical enucleation is the treatment of choice. [8,11] In large lesions, the adjacent teeth may have to be extracted because of the lack of supporting bone. In cases of lesions associated with the tooth favouring orthodontic movement, marsupialization or decompression of the lesion is recommended as the initial modality of treatment. Recurrence is very rare (0.2%) [12]

In the case reported here, all the differential diagnosis were ruled out clinicopathological correlation led to a final diagnosis of the adenomatoid odontogenic tumour. The patient was surgically treated under general anaesthesia; tumor was surgically resected, impacted canine, and lateral incisor was removed.

CONFLICTS OF INTEREST

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

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