

ADENOMATOID ODONTOGENIC TUMOR OF THE MANDIBLE — EXTRAFOLLICULAR VARIETY: A RARE CASE REPORT

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ABSTRACT

Adenomatoid odontogenic tumor (AOT) is an uncommon benign odontogenic lesion that affects young patients, with female predominance, mainly in second decade, representing approximately 3% of all odontogenic tumors. It is of interest that in about 74% of cases, this tumor is associated with an unerupted tooth, and in over two third of cases associated with maxillary (65%) or mandibular canine (35%). Very rarely the lesion occurs without any impacted tooth. In this report a case of adenomatoid odontogenic tumor of right side of mandible in the canine premolar region not associated with an impacted tooth in a 14 years old female patient is described.

Key words: Adenomatoid odontogenic tumor, association with impacted tooth, female pre-dominance

INTRODUCTION

Adenomatoid odontogenic tumor (AOT) is a relatively uncommon distinct odontogenic neoplasm that was first described by Steensland in 1905.¹ This tumor was initially labeled as 'epithelial tumor' by Stafne in 1948 and as adenoameloblastoma by Thoma in 1955. The term Adenomatoid odontogenic tumor was introduced by Philipsen and Birn in 1971. It is defined by WHO as "a tumor of odontogenic epithelium with duct like structures and with varying degree of inductive changes in the connective tissue. The tumor may be partly cystic or it may present only as a mass in the wall of the large cyst."² This tumor occurs more frequently in the anterior part of the jaws with 76% developing anterior to cuspid in the maxilla and the mandible. It has been reported that in 74% of the cases, this tumor was associated with an impacted tooth.³

CASE REPORT

A female patient aged 14 years reported to the Department of Oral Medicine and Radiology with a chief complaint of swelling on the right side of the lower jaw since three months which has gradually increased in size. There was displacement of teeth. There was no history of trauma and no relevant medical and family history. The patient was pale and her submandibular lymph nodes were enlarged, palpable, non tender and firm in consistency. Extra oral examination revealed that the face was asymmetrical with a diffused swelling on the chin, extending anteriorly from midline of the chin and posteriorly till the corner of the mouth, superio-inferiorly extending from the middle of the cheek upto the lower border of the mandible measuring approximately 2cm x 2cm in size. The swelling was firm in consistency and non tender on palpation (Fig 1a). On intraoral examination, an oval shaped swelling was

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present in the lower right canine premolar region, extending antero-posteriorly from the right canine to the second premolar, superiorly from the marginal gingiva to the vestibule inferiorly. The swelling also extended lingually from the marginal gingiva till the floor of the mouth, the size of the swelling was 2 cm x 1.5 cm and 43 was labially drifted. On palpation the swelling was firm in consistency, non tender with well defined borders (Fig 1b). Labiobuccal as well as lingual cortical plate expansion was present and 41,42,43,44 had grade I mobility. 31, 41, 42, 43, 44, 45 were all vital on electric pulp test. Considering the history and clinical examination, a clinical diagnosis of benign odontogenic tumor of the jaw was made with a differential diagnosis of ameloblastoma, adenomatoid odonto-

genic tumor, calcifying epithelial odontogenic tumor, calcifying epithelial odontogenic cyst and central giant cell granuloma.

Routine blood investigations were carried out and all the values were in normal limit except that the hemoglobin which was low suggesting that the patient was anemic. IOPA of 43, 44, 45 region showed a well defined radiolucency with small sparse pebble like calcifications seen in between the radiolucency (Fig 2a). Mandibular cross sectional occlusal projection showed labial tilting of 43 as well as buccal and lingual cortical plate expansion (Fig 2b). Panoramic radiograph revealed an oval shaped radiolucency with a well defined corticated border. Drifting of the roots of 43, 44



Fig 1a: External swelling on the chin

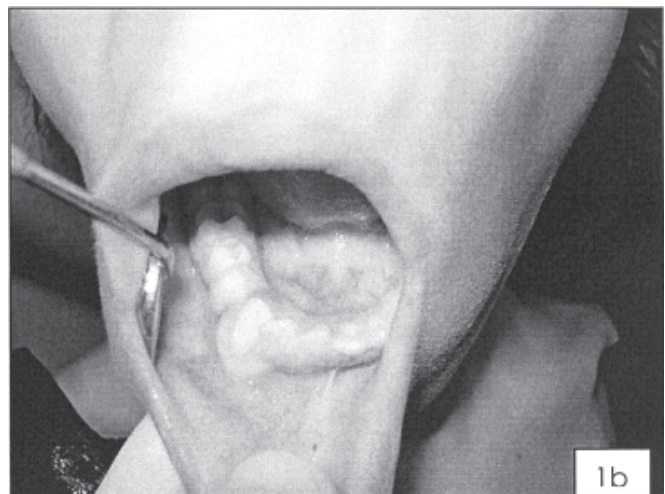


Fig 1b: Intraoral swelling extending from right canine to the second premolar

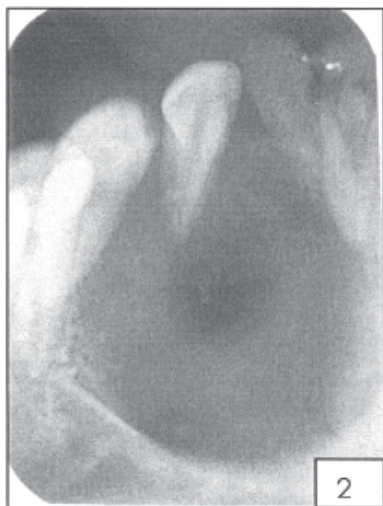


Fig 2a: Well defined radiolucent lesion with sparse pebble like calcifications

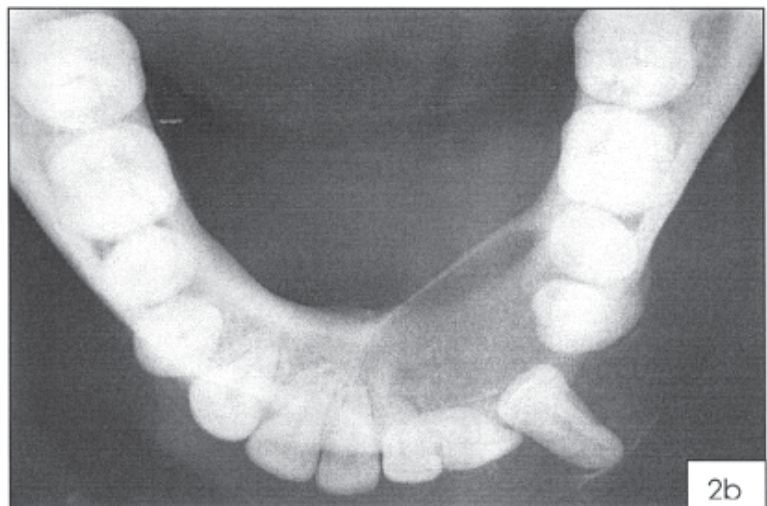


Fig 2b: Cross sectional view of the mandible showing a labially titled canine

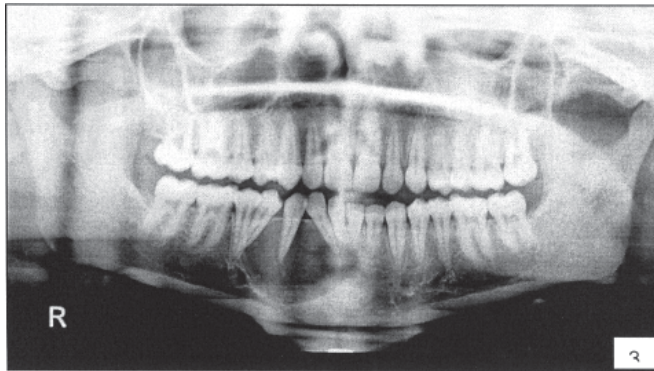


Fig 3: Well defined radiolucency and the tilted roots present in relation to 42, 43, 44



Fig 4: Axial section of CT scan showing well defined 2 x 2.2 cm size expansile unilocular lytic lesion

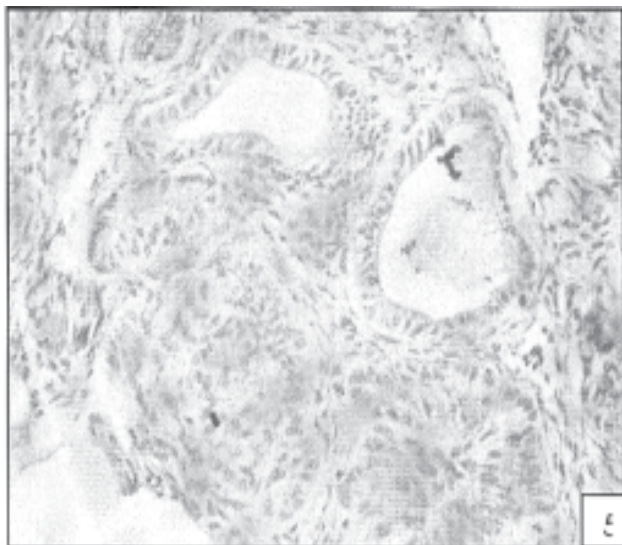


Fig 5: Nest and duct like structures lined by low columnar epithelial cells seen at 40X magnification

and 45 was seen (Fig 3). CT Scan revealed a well defined 2 x 2.2 cm size expansile unilocular lytic lesion at the right half of the mandible, incisor and premolar teeth were displaced (Fig 4).

Incisional biopsy of the lesion was performed and histopathological section showed tumor comprising of nest and duct like structures lined by low columnar epithelial cells with benign looking nuclei. Stroma comprises of spindle shaped cells. Foci of calcification were present, areas of hemorrhage were also seen and the overall picture was suggestive of adenomatoid odontogenic tumor of the mandible (Fig 5). Patient was advised for enucleation of the lesion with curettage and referred to the pediatrician for systemic management and later the patient was lost to follow up.

DISCUSSION

Adenomatoid odontogenic tumor is a slowly growing lesion, with a predilection for the anterior maxilla (ratio of cases 2:1 relative to mandible) of young females. Sixty-nine percent of adenomatoid odontogenic tumors are diagnosed in the second decade of life, and more than half occur during the teenage years. The female to male ratio for all age groups and all variants is close to 2:1. Generally the tumors do not exceed 1-3 cm in greatest diameter, but they can be larger. It commonly occurs in relation with an impacted canine, and adjacent teeth may be slightly displaced.⁴ The lesion usually presents as an asymptomatic swelling which grows slowly.¹

There are three clinicopathologic variants of AOT, namely intraosseous follicular, intraosseous extra-follicular and peripheral, all have identical histology. 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 an unerupted tooth has been reported. This type is often located between, above or superimposed upon the roots of adjacent erupted teeth. In the present case there was no impacted teeth associated with the lesion. The peripheral variant appears as a gingival fibroma or epulis attached to the labial gingiva.⁵

The origin of AOT is controversial. However, most of the authors accept its odontogenic source as it occurs in the tooth bearing areas of the jaws and in most of the cases it is found in close association with an impacted tooth. It has cytological features similar to those of various components of the enamel organ, dental lamina, reduced enamel epithelium and/or their remnants. Others believe that it is a developmental outgrowth or hamartoma owing to the limited size of most cases and lack of recurrence.⁶

The radiographic findings of AOT frequently resemble other odontogenic lesions such as dentigerous cysts, calcifying odontogenic cysts, calcifying odontogenic tumors, globulo- maxillary cysts, ameloblastomas, odontogenic keratocysts and periapical cyst. The follicular variant shows a well-circumscribed unilocular radiolucency associated with the crown and often part of the root of an unerupted tooth, the radiolucency of the extrafollicular type is located between, above or superimposed upon the roots of erupted permanent teeth.¹ Faint radiopaque foci may be seen in many cases, and some may show dense clusters of ill defined radiopacities, occasionally the calcifications are small with well defined borders, like cluster of small pebbles.⁷ Displacement of neighbouring teeth due to tumor expansion is much more common than root resorptions. The peripheral lesions may show some erosions of the adjacent cortical bone.¹ In the reported case one could appreciate faint radiopaque foci in the periapical radiograph and tilting of roots of 43, 44, 45 which is characteristic of an adenomatoid odontogenic tumor.

Histologically, the most conspicuous feature is cuboidal or columnar epithelial cells forming nests or rosette like structure with central eosinophilic amorphous material. Varying sized duct like spaces lined by low columnar cells are present within the nodules. Fragments of crystalline calcification resembling cementum may be seen. Presence of amyloid like material resembling to that of CEOT has lead some workers to propose the existence of combined AOT and CEOT. However, Montes et al concluded that CEOT like areas were within normal histopathological spectrum of AOT.² The calcified materials seen in adenomatoid odontogenic tumor have been considered to be a form of enamel, dentin, enamel and dentin, cementum, dentin and cementum or dystrophic calcifications, but their exact nature still remains a controversy.⁸

The adenomatoid odontogenic tumor is a benign tumor that presents with a non- aggressive biologic behavior, progressive growth, small frequency of recurrence, absence of invasion, and the frequent presence of a connective tissue capsule, the treatment should consist of enucleation and curettage. It should however, be noted that in cases where an impacted tooth involved by the tumor presents itself in a favorable position for orthodontic movement, with the objective of preservation of the tooth. It is recommend that marsupialization or decompression should be considered as the initial modality of treatment. However, when the tumor does not decrease in size, or continues growing during treatment by marsupialization, surgical removal should be performed.⁹

CONCLUSION

Adenomatoid odontogenic tumor is usually seen associated with an impacted tooth, mostly a canine. But the extra follicular variety, though rare also exists, so a diagnosis of adenomatoid odontogenic tumor should be considered in the differential diagnosis of corticated radiolucencies with small radiopaque foci, especially in teenagers and young adults, even in the absence of an impacted tooth.

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