

## Adenomatoid Odontogenic Tumor of Mandible – An Unusual Case Report

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### Abstract

Adenomatoid Odontogenic Tumor (AOT) is a relatively uncommon odontogenic neoplasm. Initially this lesion was considered to be a variant of Ameloblastoma but its clinical features and biological behaviour indicate that it is completely a separate entity. It is a tumor arising from odontogenic epithelium with duct like structures and varying degrees of inductive changes in the stroma. It is a slow growing benign lesion commonly found in maxillary anterior region of young females and represents 3% of all odontogenic tumors. The aim of this paper is to present a rare case of intraosseous follicular variant of AOT in the mandibular anterior region.

**Keywords:** Adenomatoid Odontogenic Tumor, Intraosseous, Follicular.

### Introduction

Adenomatoid Odontogenic Tumor (AOT) is a benign tumor originating from the odontogenic epithelium, corresponding to 2.2-7.1% of all odontogenic tumors.<sup>1</sup> The first case was reported by Steensland in 1905 as “Epithelioma Adamantinum”. Various names have been used for this lesion like “Adeno- ameloblastoma” which was used for many years since the tumor was considered a histological variant of Ameloblastoma.<sup>2</sup> The tumor was also reported by Harbitz in 1915 under the name of “Cystic Adamantoma.”<sup>3</sup> Philipsen and Birn proposed the widely accepted and currently used name “Adenomatoid Odontogenic Tumor”, a term that was adopted by the first edition of the World Health Organization classification of Odontogenic tumors in 1971. Adenomatoid Odontogenic Tumor is defined as a tumor composed of odontogenic epithelium in a

variety of histoarchitectural patterns, embedded in a mature connective tissue stroma and characterized by slow but progressive growth. AOT is mostly seen in young patients, especially in the second decade of life and is not commonly found in patients older than 30 yrs of age. Females are affected more often than males. Maxilla is the predilection site of occurrence, almost twice as often as the mandible and the anterior part of the jaw is more frequently involved than the posterior part.<sup>4</sup>

### Case Report

A 15yr old female patient reported to I.T.S Dental College, Hospital & Research Centre with a chief complaint of pain and swelling in lower front teeth region since 15-20 days. On intraoral examination, diffuse swelling was present in mandibular anterior teeth region in relation to 31, 41 and 43 with a smooth surface. Grade I mobility was present in

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relation to 31, 32, 41, 43 with missing 42 (Fig. 1). Orthopantomogram revealed a single unilocular, well defined radiolucency in the midline of the mandible with well defined borders, extending mediolaterally from apical region of 44 to 33 and extending from crest of alveolar bone till inferior border of mandible. The radiolucency completely enclosed the tooth including root of 42. Displacement of teeth was seen in relation to 31, 32, 41, 43 (Fig. 2). Occlusal radiograph showed buccal and lingual cortical plates expansion (Fig. 3). Hematological and urine analysis were done and were normal. Enucleation of the lesion was done and sent for histopathological examination.

Histopathological examination revealed odontogenic epithelial cells forming nests, nodules, whorls and duct like structures in a fibrous connective tissue stroma with hemorrhagic areas (Fig. 4). The nodular areas exhibits 2-3 layers of odontogenic epithelium with duct like structures having a single layer of odontogenic epithelium and the whorls and rosettes pattern having 2-3 layers of epithelium (Fig. 5). Areas of calcification, areas of amorphous eosinophilic material along with pleomorphic epithelial cells arranged in sheets and strands are noted. Areas of odontogenic epithelial rests are present. Histopathological examination confirmed the diagnosis of Adenomatoid Odontogenic Tumor.



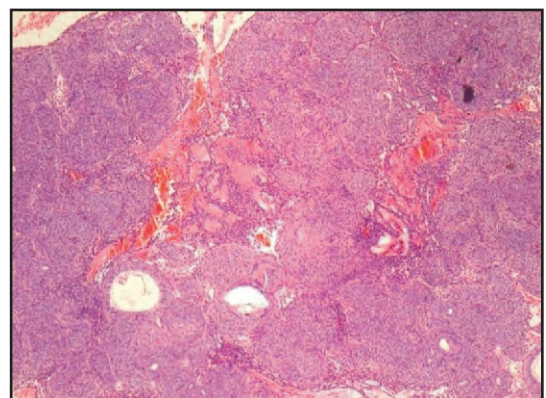
**Figure 1:** Pre-operative view showing swelling in mandibular anterior jaw region



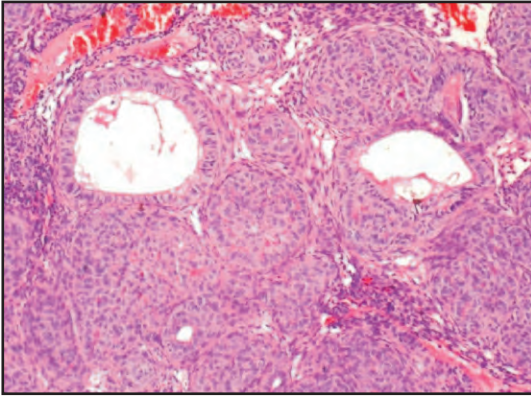
**Figure 2:** Pre-operative Panoramic view showing single, unilocular, well defined radiolucency on the midline of the mandible extending mediolaterally from apical region of 44 to 33



**Figure 3:** Occlusal radiograph shows buccal and lingual cortical plate expansion.



**Figure 4:** Photomicrograph is showing odontogenic epithelial cells forming nests, nodules, whorls and duct like structures in a fibrous connective tissue stroma with hemorrhagic areas (H & E, x40)



**Figure 5:** Photomicrograph is showing nodular areas exhibits 2-3 layers of odontogenic epithelium with duct like structures having a single layer of odontogenic epithelium and the whorls and rosettes pattern are having 2-3 layers of epithelium (H & E, x100)

**Discussion**

AOT is defined as a tumour of odontogenic epithelium with duct like structures and varying inductive changes in the connective tissue.<sup>5</sup> The tumour is largely found in the second and third decades of life with a male: female ratio of 1:1.9; 64.3% of all AOT variants occur in the Maxilla of which 62.3% are intraosseous. The canine was found to be the commonest permanent tooth to be involved.<sup>6</sup> In a study done by Swadison in Thai patients showed that out of 67 reported cases of AOT, none of the lesions was present in the mandibular region.

There are 3 clinicopathologic variants of AOT – namely intraosseous follicular, intraosseous extrafollicular and peripheral variants. The follicular type is a central intraosseous lesion associated with an impacted tooth. Intraosseous extrafollicular variant of AOT has no relation with an unerupted tooth. It is commonly located above or superimposed upon roots of adjacent erupted teeth. These 2 variants accounts for 96% of all cases of AOT. Peripheral variant appears as gingival growth on labial surface. The case

which is presented here is an intraosseous follicular variant in mandibular anterior jaw region. Radiographically, the intraosseous follicular AOT is seen as a well defined unilocular radiolucency associated with crown or root of an unerupted tooth.<sup>7</sup> The most common differential diagnosis for this variant is Dentigerous Cyst which also occurs as a pericoronal radiolucency in the jaws. Dentigerous Cyst encloses the coronal portion of an unerupted tooth in most of the cases whereas AOT encloses both coronal and radicular portion of an unerupted tooth.<sup>8</sup> In approximately two thirds of intrabony variants, the radiolucency shows discrete foci having a flocculent pattern of scattered radiopacities. In this case, the radiolucency was well defined and unilocular associated with an unerupted or impacted mandibular anterior tooth.

AOT is usually surrounded by a well developed connective tissue capsule. It may present as a solid mass, a single large cystic space or as numerous small cystic spaces. The characteristic duct like structures are lined by a single row of columnar epithelial cells, the nuclei of which are polarized away from the central lumen. The lumen may be empty or contain amorphous eosinophilic material. Dystrophic calcification is encountered in varying amounts and in different forms. It is also seen scattered among epithelial masses or in the stroma, in most AOT structures.<sup>5</sup>

Immunohistochemical and ultrastructural findings have shown that eosinophilic deposits probably represent some form of enamel matrix. Recent studies indicate that AOT is derived from a complex system of dental lamina and its remnants.<sup>6</sup> As the growth of AOT is slow growing and its nature is benign, the treatment of choice is enucleation

and curettage.

In the present case; the age, sex and histopathology is in accordance with the literature but the involved site and tooth is a rare occurrence. AOT is more common in maxilla but in this case it was present in mandibular anterior region. It is commonly associated with an impacted canine but in this case it is associated with an impacted Lateral Incisor.

### **Conclusion**

Adenomatoid Odontogenic Tumors are rare odontogenic tumors which are common in young patients. So as dentists, it is important to know about the various variants of tumor with all its features so that early diagnosis can be made properly and the treatment can be done as soon as possible.

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