CASE REPORT

EXTRAFOLLICULAR ADENOMATOID ODONTOGENIC TUMOR OF MAXILLA: A CASE REPORT

Dr. Hiral Shah [1], Dr. Neelampari Parikh [2], Dr. C.nandini[3], Dr. Anil Patel [1]

Post Graduate Student, Department of Oral Pathology and Microbiology and Forensic Odontology, Karnavati School of Dentistry.[1]Professor and Head, Department of Oral Pathology and Microbiology and Forensic Odontology, Karnavati School of Dentistry.[2]Reader, Department of Oral Pathology and Microbiology and Forensic Odontology, Karnavati School of Dentistry.[3]

Abstract

Odontogenic tumours are a group of heterogeneous lesions, features of which have been catalogued for several decades. Adenomatoid odontogenic tumor (AOT) is an uncommon, hamartomatous, benign, epithelial lesion of odontogenic origin that was first described by Driebaldt in 1907, as a pseudo-adenoameloblastoma. The current World Health Organisation (WHO) classification of odontogenic tumors defines AOT as being composed of the odontogenic epithelium in a variety of histoarchitectural patterns, embedded in mature connective tissue stroma, and characterized by slow, but progressive growth. The rarity of adenomatoid odontogenic tumor may be associated with its slowly growing pattern and symptomless behavior. Therefore, it should be distinguished from more common lesions of odontogenic origin in routine dental examinations. The current article reports a case of extrafollicular variety of adenomatoid odontogenic tumor occurred between with left maxillary canine and premoloar teeth region with divergence of roots in a 13 years old female patient.

Keyword: AOT, Extrafollicular ,Maxilla

Introduction

Adenomatoid odontogenic tumor (AOT) is an uncommon tumor.[1] It is rightfully called as master of disguise.[2] Philipsen and Birn (1969) 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.[3] WHO defined AOT as a tumor composed of odontogenic epithelium presenting a variety of histoarchitectural patterns, embedded in a mature connective tissue stroma, and characterized by slow but progressive growth.[4]

The tumor is sometimes referred to as "Two-thirds tumor" because it

occurs in the maxilla in about 2/3rd cases, about 2/3rd cases arise in young females, 2/3rdcases are associated with an unerupted tooth, and 2/3rd affected teeth are canines.[5]

In this case report, we present an interesting case of an Adenomatoid Odontogenic Tumour located in the anterior maxillary region in a 13 year old female patient.

Case Report

A 13 year old female patient reported to Karnavati School of Dentistry, Gandhinagar with the chief complaint of swelling on the left side of face since last one year. History revealed that, the swelling had gradually increased in size over a period of one year to attain the present size. There was no history of trauma in that region.

On Clinical examination, an intra oral swelling was seen on the maxillary left canine tooth region measuring approximately 1 x 1 cm in size, extending apical third region of canine eminence till the labial vestibule. The swelling was not associated with pain, discharge or paresthesia. The lesion was firm, and the mucosa over the lesion was normal without any erythematous and ulcerative changes. Both the teeth, upper left canine and upper left first premoloar were vital. Bilaterally, submandibular lymph nodes were tender and palpable. No relevant medical history was revealed.

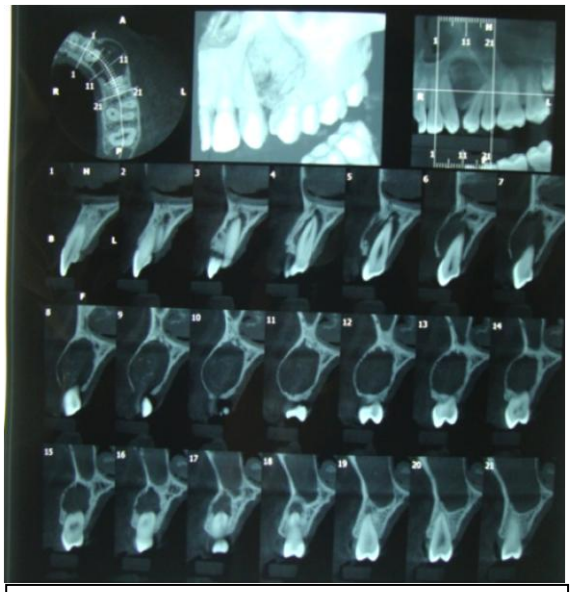

Figure 1: CBCT image showing a well defined hypodense area related to root of maxillary left canine with focal hyperdense areas within.

Radiological investigations were performed and the Cone Beam Computerized Tomography (CBCT) **(Figure: 1)** showed a well defined radiolucent area on the left side of the maxilla, extending from distal root surface of upper left canine to the mesial root surface of upper left first premoloar. The roots of both these teeth showed divergence. Depending upon the clinical and radiographic findings, a provisional diagnosis of adenomatoid odontogenic tumor was made with differential diagnosis of calcifying epithelial odontogenic cyst and dentigerous cyst.

Figure 2: Photograph of gross specimen

All laboratory investigations were carried out and found to be within normal limits before the surgical procedure. Enucleation of the lesion along with the extraction of upper left canine were performed under local anesthesia without any complication and specimen was kept in 10% formalin. The tissue specimen was sent for histopathological examination**.** **(Figure:2)** On macroscopic examination, specimen consisted of an ovoid mass of brown to grayish tissue of about 1 x 1.5 x 1.5 cm. The surface was smooth to irregular and had a firm and texture when cut. The growth was attached to the root surface of maxillary left canine. The cut surface was creamish white in color with few microcystic spaces and few hard areas. The tissue was then processed, and multiple sections were stained with hematoxylin and eosin. On microscopic examination, H & E stained sections showed a well encapsulated mass of soft connective tissue with the central part filled almost completely with tumor component. It revealed presence of

proliferating spindle-shaped epithelial cells arranged in varying pattern like whorled and rosette-like. Numerous duct-like structures of varying sizes were noted and were lined by tall columnar cells with reverse polarized nucleus. At places foci of calcification were noted. **(Figure: 3, 4, 5, & 6)**

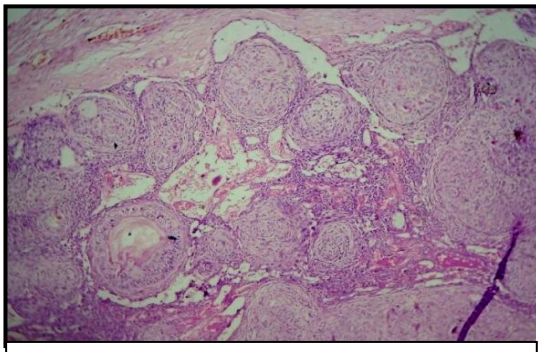

Figure: 3 H and E stain Photomicrograph showing tumor mass with fibrous connective tissue capsule (10 x)

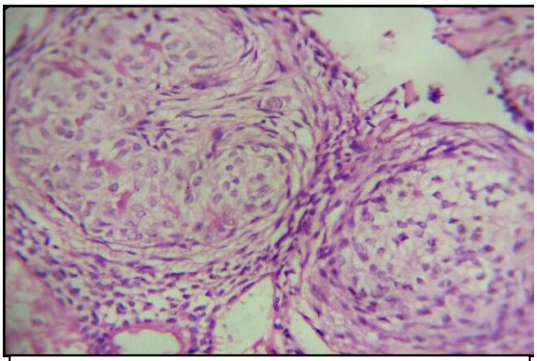

Figure: 4 H and E stain Photomicrograph showing spindle shaped cells arranged in whorls and rosette pattern (40 x)

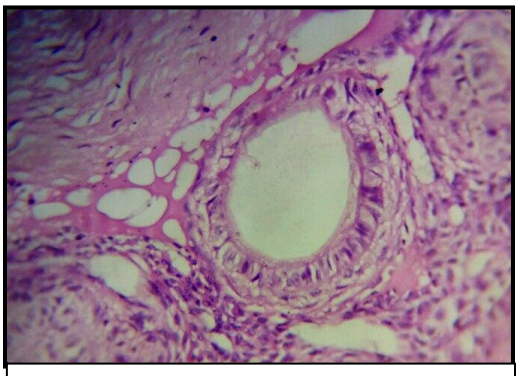

Figure: 5 H and E stain Photomicrograph showing tumors cells arranged in characteristics duct like structures lined by a single row of columnar epithelial cells.

The connective tissue stroma was scanty and showed loosely arranged collagen fibers interspersed with fibroblasts and endothelial lined blood vessels. Based on clinical, radiographic and histological findings, a final diagnosis of an Extrafollicular Adenomatoid odontogenic tumor was made.

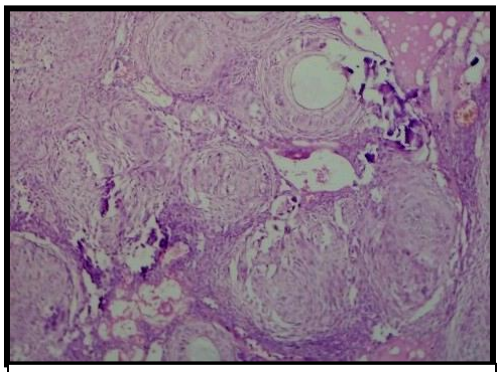

Figure: 6 H and E stain Photomicrograph showing calcification (10 x)

Discussion

Adenomatoid odontogenic tumor accounts for 2.2% to7.1% of all odontogenic tumors, which has a ranking of fourth or fifth among the odontogenic tumors.[6] More than 750 cases have been reported in the literature.[7] In the WHO classification of 2005, AOT is included under "odontogenic epithelium with mature, fibrous stroma without odontogenic ectomesenchyme". WHO defined AOT as a tumor composed of odontogenic epithelium presenting a variety of histoarchitectural patterns, embedded in a mature connective tissue stroma, and characterized by slow but progressive growth.[8]

The origin of AOT is controversial. Most authors accept its odontogenic source as it occurs within the tooth bearing areas of the jaws and is often found in close association with embedded teeth, and has cytological features similar to enamel organ, dental lamina, reduced enamel epithelium and/or their remnants suggesting it to be a neoplastic growth. Some support the idea that the lesion is a developmental outgrowth or hamartoma due to the relative size of the tumor and lack of recurrences.[4]

AOT is a benign, non invasive odontogenic tumor showing a slow growth pattern. AOT is commonly seen in young patients, especially in second decade of life. Females are more commonly affected than the males[9-11] with a ratio of 1.9:1. Maxilla is the predominant site of occurrence and twice as common than the mandible. Anterior maxilla is more commonly affected than the posterior part.[8] These features coincide with the case presented in our study. The patient of the present case was a female aged 13 years. The tumor was located in anterior maxillary region.

Majority of the lesions are asymptomatic and measured between

1.5 to 3 cm, although larger lesions can exceed 7 cm. While peripheral lesion appears as gingival color mass ranging from 1 to 1.5 cm.[1] In our case size of lesion was almost 1.5 cm. The main types of AOT include follicular, extrafollicular, and peripheral with the follicular being most common and makes up 70% of the cases.[4, 12] In about 75% of cases, the follicular variant appear as well‑defined unilocular (round or ovoid) radiolucency associated with a crown of unerupted tooth, most often canine. Whereas the radiolucency of the extrafollicular type is located between, above or superimposed upon the roots of erupted permanent teeth.[6,13,8] As it is seen in our case. The peripheral type may show slight erosion of the alveolar bone crest, but radiographic changes are often difficult to detect. Displacement of neighboring teeth due to tumor expansion is much more common than root resorption. The patient described in this report presented no root resorption, but showed divergence of the roots of maxillary left canine and maxillary left premolar. It also contains a minimal amount of radiopacities described as flecks, snowflake. As Dare et al. (1994) suggested that intraoral periapical radiographs are superior to orthopantomographs in detecting the characteristic radiopacities.[6] Konouchi et al. (2000) suggested that magnetic resonance imaging was useful to distinguish AOT from other lesions, even if it is difficult on periapical radiographs.[14]

AOT is usually surrounded by a well‑developed connective tissue capsule while Garg et al. (2009) have reported a case fast growing AOT which was unencapsulated and associated with root resorption.[2] It may appear either as a solid mass, a single large cystic space or as numerous small cystic spaces. The tumor is composed of spindle‑shaped or polygonal cells forming sheets and whorled masses in a scant connective tissue stroma. Between the epithelial

cells, as well as in the center of the rosette-like structures, are amorphous eosinophilic materials. The characteristic duct-like structures are lined by a single row of cuboidal to columnar epithelial cells, the nuclei of which are polarized away from the central lumen. These duct-like structures are frequently lined by eosinophilic rim of varying thickness the so-called "hyaline ring". Dystrophic calcification in varying amounts and in different forms like irregular masses, Liesegang rings, spheroidal and globular masses is usually encountered in most AOTs within the lumina of the duct-like structures or scattered among epithelial masses or in the loose, hypocellular, fibrovascular stroma.[1,2,4] In this case, all this classical features were predominantly revealed.

Immunohistochemically, Larson et al. (2003) characterized the classical AOT by a CK profile similar to follicular cyst and or oral or gingival epithelium based on positive staining with CK5, CK17 and CK19. Crivelini et al. (2003) detected the expression of CK14 in AOT and concluded its origin from reduced dental epithelium that was also positive to staining with CK14 antibodies. Abiko et al. (2001) showed positive reactions for amelogenin in limited areas in AOT.[14]

Conservative surgical enucleation is the treatment modality of choice because of its capsule. Recurrence of AOT is rare. Only three cases in Japanese patients are reported in which the recurrence of this tumour occurred; therefore, the prognosis is excellent when the lesion is completely removed.[3]

Conclusion

It should be emphasized that extrafollicular variant of AOT is very rare. Only careful diagnosis and adequate interpretation of clinical

and radiographic findings may be helpful in arriving at a correct diagnosis; however, the treatment plan remains the same. CBCT can be effective tool to find out the extension and character of the lesion.

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Corresponding author:
Dr. Hiral S.Shah.20-Shitalkunj Society, Jawahar chowk, Sabarmati, Ahmedabad.
Email: drhiralsshah@gmail.com, Conflict of Interest: None, Financial Support: None