CASE REPORT

Extrafollicular Variant of Adenomatoid Odontogenic Tumor in Maxilla: A Diagnostic Enigma

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Abstract

Introduction: Adenomatoid odontogenic tumor (AOT) is a rare epithelial odontogenic tumor of jaws. It is a benign, painless, non-invasive, slow-growing lesion, with a frequency of 2.2-7.1%. Upon clinical examination it can be misdiagnosed as an odontogenic cyst. AOT affects young individuals with a female predominance, occurs mainly in the second decade, more common in anterior maxilla and usually surrounds the crown of unerupted tooth mostly impacted canine. AOT has three clinical subtypes named as follicular, extrafollicular, and peripheral.

Clinical presentation: This case report represents a 20 years old female patient with the chief complaint of swelling on the right upper anterior region for 8 months.

Management and prognosis: After surgical excision and histopathological evaluation it is diagnosed as a rare case of extrafollicular variant of intraosseous AOT.

Key words: Adenomatoid, extrafollicular, Odontogenic Tumor

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Introduction

Adenomatoid odontogenic tumor (AOT) was first described in 1915 by Harbitzas as cystic adamantoma[1]. Philipsen and Birn proposed the name adenomatoid odontogenic tumor, whichwas adopted by the World Health Organization classification (WHO) of odontogenic tumorsin 1971[2]. WHO (2005) 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"[3]. AOT is an uncommon benign odontogenic tumor and it has an occurrence rate of 2.2 - 7.1%[4]. It appears as an intraoral-extraoral swelling and is referred to as "Two-thirds tumor" because it occurs inmaxilla about 2/3rd cases, arises in young females about 2/3rd cases, 2/3rd of the cases are associated with an unerupted tooth and among them 2/3rd affected teeth are canines[5]. Currently, it is generally accepted to be a true neoplasm rather than a hamartoma.

Case Report

A 20-year-old woman presented with the chief complaint of swelling on the right upper front teeth region since 8 months. The swelling was small in size when first noticed and gradually increasing to obtain the present size. There was no pain, tenderness, bleeding or discharge associated with the swelling.

Extraorally, the swelling was located in the lower third of the face on the right side lateral to the nose, round in shape and measured approximately 2.5x2 cm, extending superiorly up to the nasolabial fold, laterally to the zygomatic bone, and inferiorly to the corner of the mouth. (Fig. 1) On palpation, the swelling was nontender, immobile and bony hard in consistency. Intraorally, the swelling was present in the buccal vestibule on the right side, measuring 3 x 3 cm, oval in shape, extending from the distal surface of 12 to the distal surface of 14 and covering the 3/4th portion of 13, which was lineally displaced. The overlying mucosa was normal with obliteration of buccal vestibule. (Fig. 2) On palpation the swelling was nontender and firm in consistency, grade I mobility of 13 was noted.

Intra Oral Periapical Radiograph of 13,14 region showed a well defined unilocular radiolucency in the periradicular and interdental region, size approx 2.5 x 2 cm with a thin corticated continuous border. A periapical radiolucency is also seen

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in relation with 13. (Fig. 3) Maxillary cross-sectional occlusal radiograph shows expansion of buccal cortical plate and some small calcified specks like structures. (Fig. 4)

The provisional diagnosis was made as odontogenic cyst involving 13,14 region and the differential diagnosis was dentigerous cyst.

After the surgical excision of the complete lesion with involving tooth 13, the sample was sent to Department of Oral Pathology for histopathological evaluation. (Fig. 5) The specimen was encapsulated and the cut surface was homogenenous tan-white in appearance. (Fig. 6) Section stained with Haematoxyline and Eosin (H&E) showed well-circumscribed thickened fibrous capsule which

Fig. 1 Extraoral swelling

Fig. 2: Intraoral swelling

Fig. 3: Intra Oral Periapical Radiograph showing periradicular and interdental radiolucency

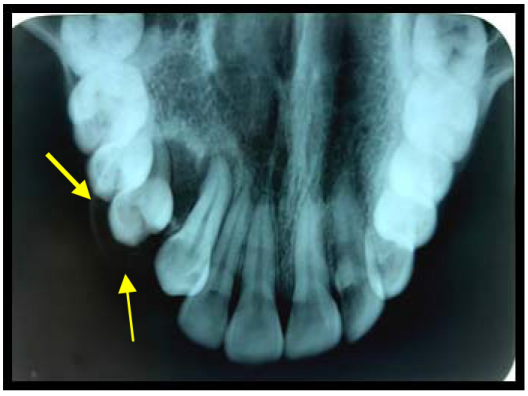

Fig. 4: Maxillary cross-sectional occlusal radiograph showing expansion of buccal cortical plate

Fig. 5: Per operative finding shows presence of well defined firm mass involving buccal vestibule of 13,14 region

Fig. 6: Gross specimen measuring about 3.0 x 2.0 x 1.0 cm3

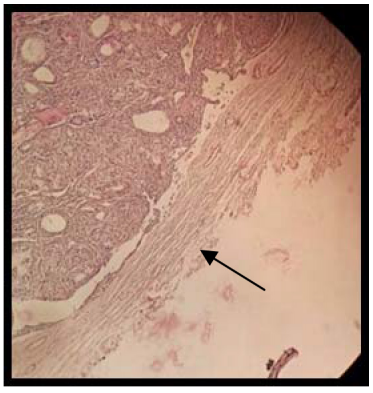

Fig. 8: Multiple rosette pattern and duct like structures composed of odontogenic epithelial cells (H&E 10x)

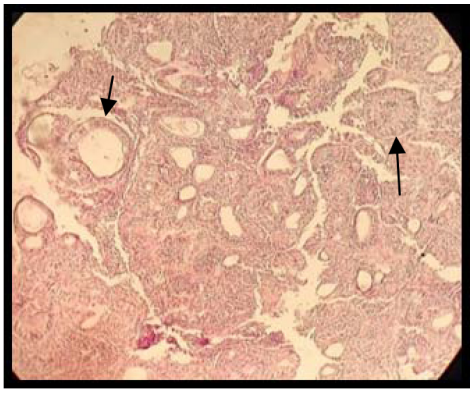

Fig. 7: Thick fibrous capsule surrounding the tumor mass (H&E 10x)

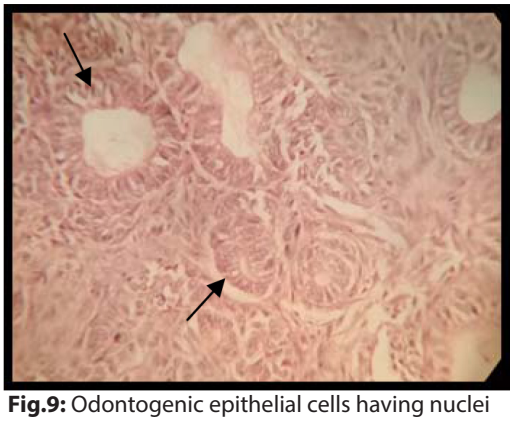

Fig.9: Odontogenic epithelial cells having nuclei towards the basement membrane and away from the luminal surface (H&E 40x)

surrounds areas of odontogenic epithelium arranged in multiple circular rosette and duct-like pattern. Few areas of eosinophilic, amorphous, amyloid-like substances were noted in the intercellular spaces. Nuclei in the columnar odontogenic epithelial cells of the ductal and rosette pattern are situated away from luminal surface without any atypia, which is suggestive of Adenomatoid Odontogenic Tumor involving 13 region. (Figs. 7 & 8)

Correlating all the history, clinical and radiological findings along with histopathological features it was concluded as a case of Extrafollicular variant of Intraosseous Adenomatoid Odontogenic Tumor involving Maxillary anterior region.

Discussion

AOT is a benign, non-invasive slow growing painless odontogenic lesion which is generally intraosseous, but can also occur rarely in peripheral locations. AOT is mostly occured in young individuals, especially in the second-third decade of life. Females are affected more often than males with a ratio of 1.9:1. The highest female incidence ratio are being observed in Sri Lanka (3.2:1) and Japan (3:1)[6].

The origin of AOT is controversial. However, most authors accept its odontogenic source. It is seen within the tooth-bearing areas, associated with embedded teeth and the histological features are similar to various components of the enamel organ, dental lamina, reduced enamel epithelium and their remnants.

The maxilla is almost twice as frequent as that in the mandible, and the anterior part of the jaw is more frequently involved than the posterior part and unerupted maxillary canine is most commonly associated with AOT[4]. Irregular root resorption is seldom reported with displacement of tooth.[7] Because of the slow growing nature of the lesion, the patients do not have any complain for years until it produces an obvious deformity. Delayed eruption of a permanent tooth or a regional swelling of the jaws may be the first symptom noted and it is frequently discovered during routine dental check up and radiographic examination[8].

Intraosseous AOT is divided into two types depending on their radiological features such as; follicular (or pericoronal) and extrafollicular (or extracoronal). The follicular variety is characterized as a well defined unilocular radiolucent lesion surrounding the crown, and is often part of the root of an unerupted tooth. The extrafollicular variety represents a well-defined radiolucent lesion, located between, above, or superimposed upon the root of an unerupted tooth. There are minute, variable-shaped radiopacities, frequently found within the lesion. The extraosseous or peripheral variety of AOT is represented a slight erosion of the underlying alveolar bone cortex.[9]

Conclusion

The case discussed emphasizes the importance of recognizing odontogenic neoplasms in oral cavity. AOT hasunique clinical, radiological, and cytological features. However, the clinical and radiographic features may often present mimics odontogenic cysts. That's why persistence of deciduous teeth for a longer duration and/or unerupted succeeding permanent teeth, when associated with a swelling, always need to be investigated thoroughly.

Declaration of patient consent

The authors certify to obtain all the appropriate consent forms from the patient for her images and other clinical information to be reported in the journal.

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