

Adenomatoid Odontogenic Tumor in the Mandible: A Case Report

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Abstract Adenomatoid odontogenic tumor (AOT) is a rare odontogenic tumor often misdiagnosed as odontogenic cyst. AOT is predominantly found in female patients, which usually arise in the second or third decade and is located more often in the maxilla than mandible and often associated with an unerupted permanent tooth. However, AOT frequently resembles other odontogenic lesions such as dentigerous cysts or ameloblastoma. Treatment is conservative and the prognosis is excellent. For illustration a rare case of an AOT in the mandible which is associated with an unerupted permanent canine is presented.

Keywords: extra follicular, adenomatoid odontogenic tumor, AOT- Mandible

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1. Introduction

Adenomatoid odontogenic tumor (AOT) are rare, slow growing, benign, odontogenic, epithelial tumors which usually arise in the second or third decade. (Philipsen H.P et al, 1991). AOTs are usually located in the anterior region of the maxilla; they normally, produce slow swelling without pain (Toida, M et al 1990). This tumor growth will cause displacement of the teeth rather than root resorption. (Engin, B et al, 2001) (Bravo M et al, 2005). Philipsen and Birn proposed the name adenomatoid odontogenic tumor in 1969 and suggested that it not be regarded as a variant of ameloblastoma because of its different behaviour (Philipsen HP et al, 1969).

Adenomatoid odontogenic tumor grouped into 3 variants (Regezi JA et al, 1978) (Courtney RM et al, 1975) the follicular type (accounting for 73% of cases), which has a central lesion associated with an embedded tooth; the extra follicular type (24% of case), which has a central lesion and no connection with the tooth; and the peripheral variety (3% of cases). This report describes a follicular adenomatoid odontogenic tumor in the mandible, illustrates the clinical, microscopic and biological features of the tumor and emphasizes the importance of the relation between the dental follicle and the tumor tissue.

2. Case Report

A 29-year-old female patient reported to our dental clinic with a chief complaint of pain and swelling over left side of lower jaw of 1month duration (Figure 1). On examination of the patient, there was a diffuse extra-oral swelling in the mandibular anterior region. The patient

had no systemic diseases nor used any medication. There was no history of trauma, pain, discharge or any other symptoms related to the lesion. In the clinical examination of the head and neck, chronic lymphadenopathy was found on the right and left submandibular lymph nodes. There was no paraesthesia over the mental region. Intraorally, the patient presented a painless, slowly increasing swelling in the mandibular anterior region. A mild labiolingual expansion was seen on the mandibular anterior alveolus to teeth 34 to 42. The tooth 33 was missing. Mucosa overlying the swelling was normal. Fine needle aspiration yielded no fluid. A OPG revealed a well-circumscribed radiolucent lesion with a radiopaque foci, and loss of cortical bone with an impacted permanent canine (Figure 2). Root resorption of incisors, canine and premolars is seen in the third quadrant. On the basis of the clinical and radiographic findings, the differential diagnosis was adenomatoid odontogenic tumor, calcifying odontogenic cyst, ameloblastic fibrous odontoma, calcifying epithelial odontogenic tumor, infected dentigerous cyst, and unicystic ameloblastoma.



Figure 1. A diffuse extra-oral swelling in the mandibular anterior region



Figure 2. A CT in the axial view



Figure 3. Surgical site

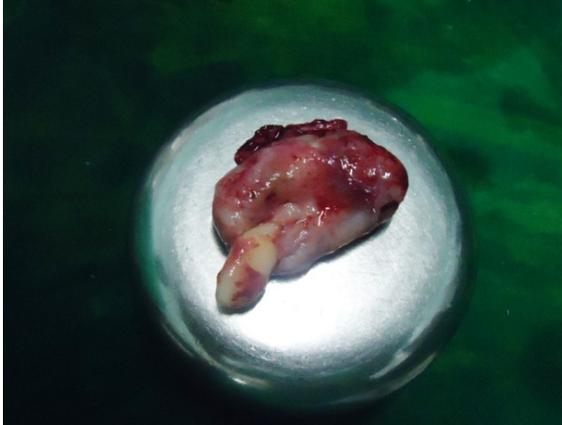


Figure 4. 22 x 18 mm tumor with the impacted canine



Figure 5. Post Operative Pic. With Vaccum drain

The patient underwent surgery with local anaesthesia. A mucoperiosteal flap in the left canine region was reflected to expose the labial aspect of the tumor. The labial cortex was very thin and had several areas of complete resorption. The tumor was totally enucleated along with the impacted lower permanent canine (Figure 3 & Figure 4). The areas between the roots of the involved teeth were curetted well a suction drain was placed and the flap was sutured in place (Figure 5). Healing was uneventful, and the suction drain was removed after three day.

3. Histopathological Findings

The lesion was a well defined mass 22x18 mm in size that was surrounded by a thick, fibrous capsule. The mandibular canine was encapsulated. After the specimen was fixed with 10% formalin, paraffin sections were prepared for light microscopy and stained with hematoxylin and eosin using routine methods. Histological examination revealed amorphous eosinophilic droplets and foci of dystrophic calcification scattered in and around the epithelial elements (Figure 7). The epithelial elements consisted of polyhedral cells in loose and irregular arrangements, spindle cells, and tall columnar cells that formed duct like structures as a result of the degeneration of the stromal tissue. The histopathological report confirmed the diagnosis of adenomatoid odontogenic tumor.

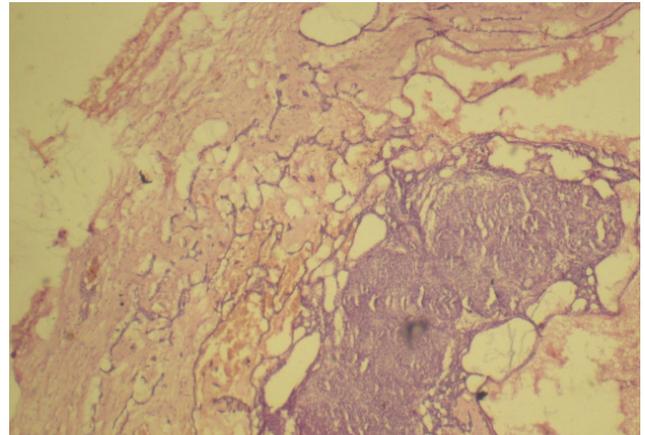


Figure 6. Scanner view showing a thick capsule and darkly stained spindle shaped cells

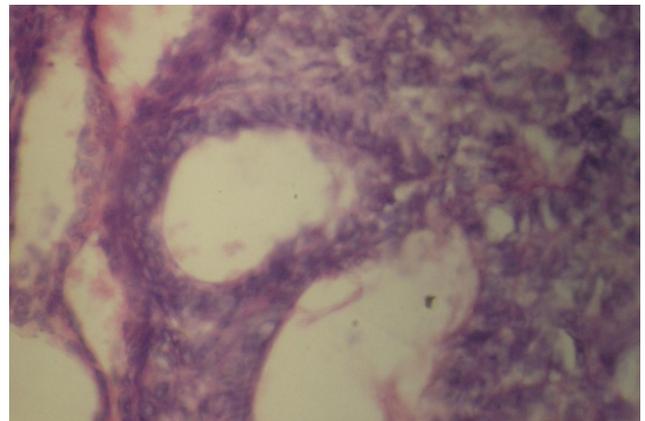


Figure 7. 10x view showing a duct like areas with tall columnar cells

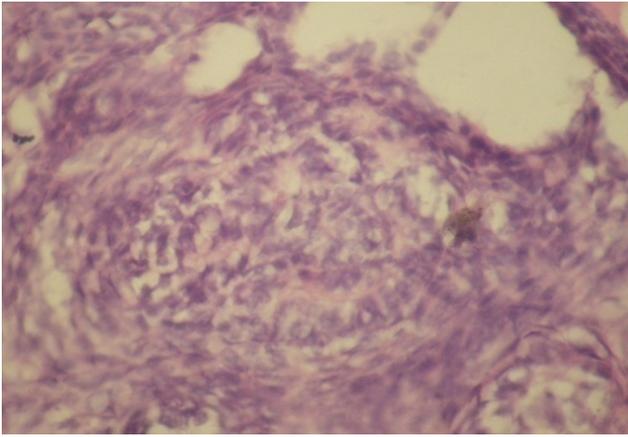


Figure 8. 40 x view showing whorl and rosette formation

4. Discussion

Adenomatoid odontogenic tumor is a slow growing lesion, with a predilection for the anterior maxilla (ratio of cases 2:1 relative to mandible) of young females. They are diagnosed in the second decade of life, and more than half occur during the teenage years. The female: male ratio is 2.3: 1 (Philipsen HP et al, 2002) (Chattopadhyay A et al 1994). The AOT was predominantly found in the upper jaw (maxilla: mandible = 2.6: 1). The lesions are typically asymptomatic, but growth of the types with central lesion results in cortical expansion. The teeth involved are generally impacted, and the adjacent teeth may be slightly displaced (Geist SY et al, 1995). It can cause a painless hard swelling, as in the case reported here and can be found on routine radiographic examination or Computed tomography. Adenomatoid odontogenic tumors, accounting for approximately 3% of all odontogenic tumors, are less frequent than odontoma, cementoma, myxoma and ameloblastoma. It has been suggested that this tumor may be a hamartoma rather than a true neoplasm (Regezi JA et al, 1978), but there is currently no evidence to resolve this dispute. For cases in which the lesion appears to surround an unerupted tooth and has no radiopaque component, dentigerous cyst may also be considered in the differential diagnosis. However, an adenomatoid odontogenic tumor often appears to envelop the crown as well as the root, whereas dentigerous cysts do not envelop the roots (Curran AE et al, 1997) (Lee JK et al, 2000).

The origin of adenomatoid odontogenic tumors is controversial (Tajima Y et al, 1992) (Philipsen HP et al, 1996) (Philipsen HP et al, 1997) some believe they originate from the odontogenic epithelium of a dentigerous cyst. In addition to the anterior maxilla, the tumor has been reported in other areas of the jaw, such as the angle of the mandible. Therefore, dental lamina remnants likely represent the progenitor cells for this benign odontogenic tumor. According to this hypothesis, the lesion grows (sometimes while forming a cystic space) next to or into a nearby dental follicle, leading to the “envelopmental theory” (Philipsen HP et al, 1992). In the case reported here, the lesion surrounded a fully formed canine, which suggests “envelopmental” pathogenesis.

Recent reports indicate that the cells of an adenomatoid odontogenic tumor usually differentiate toward an

apparent ameloblastic phenotype but fail to achieve further functional maturation (Philipsen HP et al, 1998).

Since all variants show identical benign biological behaviour and almost all are encapsulated, conservative surgical enucleation or curettage is the treatment of choice. Recurrence has been reported in very few cases (Philipsen HP et al, 1997).

5. Summary

The present case is a rare report of an adenomatoid odontogenic tumor presented in mandible of a 29 year old female involving an impacted canine. Additionally, it supports the above mentioned general description of AOT to the previous studies. Therefore, it should be distinguished from more common lesions of odontogenic origin in our routine dental examinations.

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