

Case Report

ADENOMATOID ODONTOGENIC TUMOR. CASE REPORT

Zohair Haidar, DDS, MSc, FDSRCS*

يعتبر الورم شبيه الغدي ذو المنشأ السني أنه نادره جداً تصيب أحد الفكين العلوي أو السفلي .
 تم في السنوات العشر الأخيرة تشخيص ٤ حالات من هذا الورم في كلية طب الأسنان بجامعة الملك سعود .
 نقدم في هذا التقرير حالة خامسة مع مراجعة لما كتب عن هذا الموضوع في الأدب الطبي .
 شابهت هذه الحالة في موصفاتهما كيساً تاجياً في الفك السفلي .
 تم أيضاً مناقشة طريقة التعامل مع حالات كهذه .

Adenomatoid odontogenic tumor is a very rare tumor that affects the upper or lower jaws. In the past 10 ten years only four such tumors were diagnosed at the College of Dentistry, King Saud University. A fifth case is presented with a review of the literature. The current case appeared to mimic a dentigerous cyst in the mandible. The management of such cases is also discussed.

Introduction

Adenomatoid odontogenic tumor was classified in many reports as a variant of ameloblastoma, but Stafne¹ in 1948 considered it as a distinct entity. The first description of the tumor was documented in 1907 and was called as a pseudo-adenoma adamantinoma.² In 1969, Philipsen and Birn³ proposed the name adenomatoid odontogenic tumor and, due to its different behavior, suggested that it should not be regarded as a variant of ameloblastoma. Since that time, much evidence has reinforced this concept.

Our current understanding of the odontogenic adenomatoid tumor has come about through analyzing the information gained from numerous case reports. Several extensive review articles dealing

with the subject is available.^{4,10} A survey was compiled by Courtney et al⁴ in 1975 where twenty cases had been studied. In 1970, Giansanti and colleagues⁵ investigated 108 reported cases and added three cases to the literature.

Case Report

A 20-year-old male Saudi university student was referred to the Oral Surgery Division to investigate a swelling in the chin region. The patient also noticed tilting in some anterior lower teeth sometime before observing the swelling. However, no pain was reported.

On examination, the patient looked healthy with an obvious swelling over the left side of the chin but no cervical lymphadenopathy [Fig. 1] was noticed. Mouth opening was normal, with no deviation on maximum opening.

Intraorally, the patient had almost full complement of teeth except for a missing lower left

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* Associate Professor & Consultant, Department of Maxillofacial Dental Sciences, King Saud University College of Dentistry, PO Box 60169, Riyadh 11545, Saudi Arabia



Figure 1. Photograph showing the patient with extra-oral swelling affecting the left side of the mandible.



Figure 2. Photograph showing the intra-oral view of the affected region.

canine and a retained lower deciduous left canine. The overlying mucosa appeared normal in color and texture. A firm buccal swelling extending from the lower right central incisor to the region of the retained deciduous left lower canine could be seen [Fig. 2]. There was also lingual expansion in

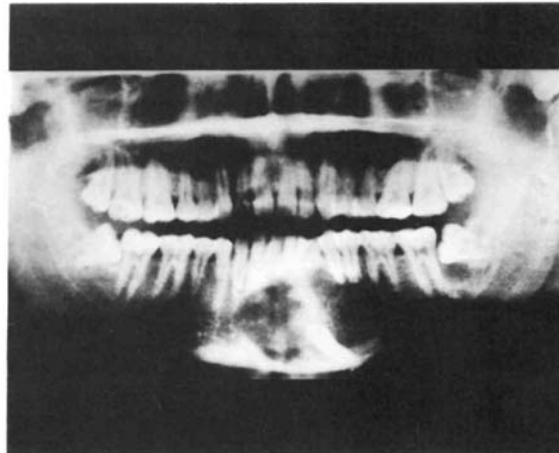


Figure 3. An orthopantomogram showing the lesion associated with impacted lower canine.

the lower left incisor-canine region. A tilting of the teeth, associated with swelling, could be seen with no abnormal mobility of any of them. All lower teeth reacted positively to thermal and electric pulp testing. No alteration in lip sensation was recorded. An orthopantomogram revealed a large, well-circumscribed radiolucency extending throughout the anterior part of the mandible from the right to the left premolar regions [Fig. 3]. The lesion expanded, with resorption of the inferior cortex in some area, extending upward into the alveolar process disturbing the usual orientation of the lower anterior teeth. The unerupted permanent lower left canine was associated with the lesion. Differential diagnoses of the lesion included odontogenic cyst and odontogenic tumor.

The patient's general health was excellent and examination of his systems, as well as his laboratory results, were within normal limits.

Under endotracheal anesthesia, a labial mucoperiosteal flap, extending from the right to the left second premolar region, was raised. The buccal plate in the anterior part of the mandible was found to be very thin and, in some areas, eroded. A tumor mass was found occupying the



Figure 4. The lesion appears following raising of the buccal flap.

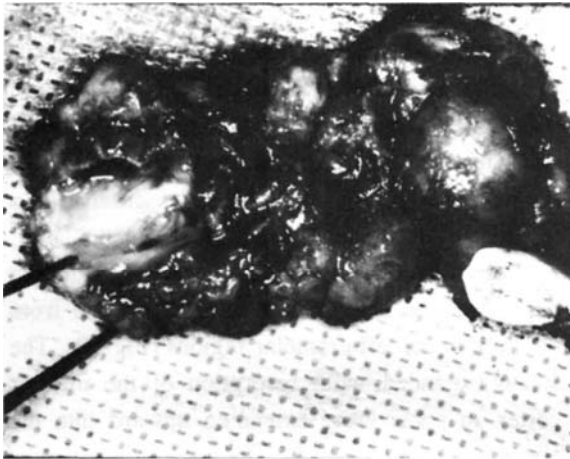


Figure 5. Gross appearance of the enucleated lesion with attached tooth.

whole chin region [Fig. 4] and was enucleated together with the attached unerupted lower left canine [Fig. 5], The lingual plate was found to be perforated in one area.

None of the anterior teeth was attached to the tumor mass and were, therefore, left in position to be clinically followed post-operatively. The specimen, including the canine tooth, was histologically examined. The resultant bony defect was left to fill in with the patient own blood. A Redi-Vac drain was inserted before suturing the buccal flap

back into position. His post-operative recovery was uneventful and healing of the surgery site was normal.

Histopathology

Microscopic examination described multiple sections from different planes of the specimen received which were exhaustively stained with H&E and trichrome stains. Histology revealed a very interesting lesion because of the juxtaposition of the odontogenic epithelial and fibrous connective tissues.

The odontogenic epithelium, which is lesional, consisted of spindle and cuboidal or columnar cells without clear cell boundary. These cells were arranged in whorls, sheets, organoid and alveolar patterns, or frank ducts. In between cells, basophilic calcified amorphous masses were present. These calcific masses were also present next to, but at times outside, the adenomatoid arrangement of the cells [Figs. 6,7].

The other areas of the section histologically showed desmoplasia with whorling and dense collagenization of the connective tissues. Normal mitoses were evident. The border of the lesion was present corresponding to the attenuated buccal

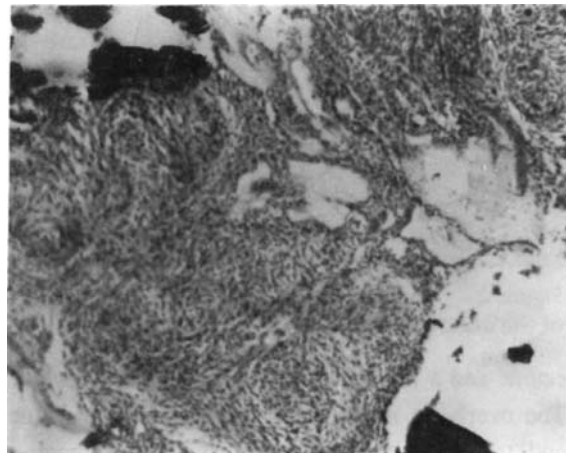


Figure 6. Adenomatoid odontogenic tumor. A cellular part of the lesion showing whorls and strands of epithelial cells but also containing amorphous calcifications.

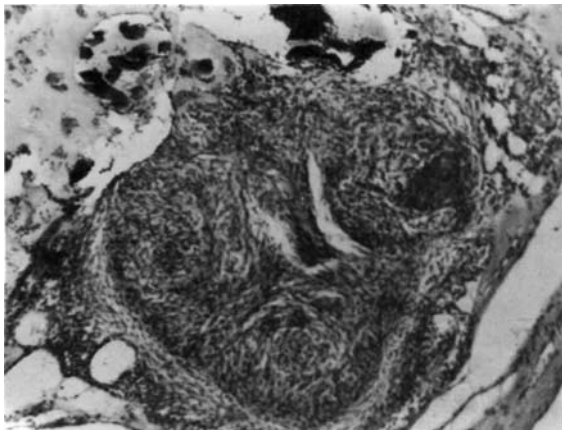


Figure 7. Whorled masses of epithelial cells in solid formations with evidence of duct-like microcysts formation at the lower left-hand corner. Amorphous calcifications are also evident. Other areas of the lesion (not shown) showed a well-defined fibrous capsule. (H&E, original magnification x100)

and lingual plates, where bone trabeculae undergone resorption. Dysplastic bone appeared to be forming in the aggressively cellular area of the desmoplastic zones. The extensive fibromyxoid tissue configures as a very broad connective tissue capsule of partly ductal and partly cystic but predominantly solid, odontogenic epithelial lesion. Diagnosis was adenomatoid odontogenic tumor, with unusually extensive fibromyxoid connective tissue.

Clinical Course

Due to the benign nature of the tumor, a decision was made to observe and follow-up the operating site regularly at 6-month intervals. All teeth near the lesion reacted positively to vitality testing methods.

Discussion

The adenomatoid odontogenic tumor is an uncommon histological type of odontogenic tumor which is characterized by the formation of duct-like or rosette-like structures by the epithelial component. Recent ultrastructural and enzyme - histochemical studies suggest that adenomatoid odontogenic tumor originates from the enamel organ epithelium.¹²

Table 1. The investigated photo-cured glass ionomer restoratives.

Age (years)	Sex	Site	Classification
30	Female	mandible	adenomatoid odontogenic tumor
21	Female	mandible	adenomatoid odontogenic tumor
35	Male	mandible	adenomatoid odontogenic tumor developed from the lumen of the calcifying odontogenic cyst
27	Female	maxilla	adenomatoid odontogenic tumor

When compared to the simple ameloblastoma, the odontogenic adenomatoid tumor has different clinical features and microscopic configuration. It is a lesion which characteristically affects the anterior part of the jaws of patients in their second decade of life.⁵ It presents as a slow-growing, painless swelling. Radiographically, the odontogenic adenomatoid tumor is seen as a unilocular radiolucency that is frequently associated with an unerupted tooth. Conservative surgical excision is curative. The ameloblastoma on the other hand is most frequently noted in the posterior part of the mandible of patients in the fourth decade. Radiographically, ameloblastoma tends to present as a multilocular lesion rather than the dentigerous cyst-like radiolucency associated with the odontogenic adenomatoid tumor. Ameloblastoma tends to be locally invasive necessitating more radical procedure.

A review of the records at the College of Dentistry, King Saud University in Riyadh showed that in the past ten years only four cases of adenomatoid odontogenic tumor were diagnosed (Table 1).

In our case, the patient was male and the affected region was the mandible. 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, the adenomatoid odontogenic tumor often appears to envelop the crown and root as in this case. Dentigerous cysts rarely encircle the root.⁵ Enucleation of those lesions should be sufficient to effect treatment. The reported recurrence rate of adenomatoid odontogenic tumor is very low (0.2%).⁸¹³

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