

A case of extrafollicular adenomatoid odontogenic tumor with long-term follow-up

Cennet Neslihan Eroglu, Serap Keskin Tunc, Omer Gunhan

ABSTRACT

Introduction: Adenomatoid odontogenic tumor (AOT) is a benign, hamartomatous, slow-growing lesion. The lesion, which is usually located in the anterior maxilla and is more prevalent in female patients, can be classified into three subgroups: follicular, extrafollicular, and peripheral. **Case Report:** This paper discusses a case of a 19-year-old female presented with swelling of the anterior maxilla, and was diagnosed with extrafollicular adenomatoid odontogenic tumor. **Long-term follow-up** was carried out after surgical treatment of the lesion. **Conclusion:** In case of the tumor contained cystic structures on histopathologic examination, long-term follow-up should be better to screen for possible recurrence of the tumor.

Keywords: Adenomatoid odontogenic tumor, Anterior maxilla, Cystic, Extrafollicular

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INTRODUCTION

Adenomatoid odontogenic tumor (AOT) is a rare, painless, benign lesion seen in young people (20–30 years of age). It usually involves the anterior maxilla and is associated with canine teeth. Overall, it accounts for 3–7% of odontogenic tumors [1–3].

The lesion was named pseudo-adenameloblastoma in 1905, but despite initial differences in nomenclature, it was recognized by Staphne in 1948 that the lesion shows different pathological structures [4–6].

Three subgroups have been defined for AOT: follicular, extrafollicular, and peripheral [7]. The follicular type accounts for 73% of AOT cases and is associated with an unerupted tooth. The extrafollicular type is not associated with an impacted tooth, and accounts for 24% of AOT cases. The peripheral type accounts for 3% of cases, and affects the gingival mucosa [8]. AOT is two times more prevalent in women than in men [9].

Adenomatoid odontogenic tumors grow slowly, cause bone expansion and tooth displacement [10]. Tumor can reach huge sizes and lead to serious deformities [11].

This case report discusses the clinical features of extrafollicular-type AOT located in the anterior maxillo-palatine region.

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CASE REPORT

A 19-year-old female patient presented with swelling in the right superior buccal and palatine regions. Her preliminary case history revealed no systemic disease or history of trauma. Intraoral examination revealed a hard swelling, extending from the right first premolar to the left first central tooth, with a surface similar to the surrounding mucosa and causing expansion of the buccal and palatine cortex, and mobility of the adjacent teeth (Figure 1). On extraoral examination, the nasobuccal sulcus was indistinct and asymmetric, with no lymphadenopathy detected. Radiological examination detected a unilocular radiolucent lesion with definite margins, causing migration of the adjacent dental roots and resorption of the root of the right superior central tooth, pushing the impacted tooth far from its normal location. We were unable to obtain fluid via aspiration, which is performed to obtain additional information about lesion characteristics. The patient underwent surgical intervention under local anesthesia, and the mass was completely enucleated. A supernumerary tooth and a persistent deciduous canine tooth within the region were extracted at the same time, but the patient did not provide consent for the extraction of the impacted canine tooth. That was seen lesion borders were not in contact with impacted tooth or follicle while the enucleation of tumor. Enucleated material was sent for histopathological examination (Figure 2). This revealed tumor particles comprising solid islets consisting of preameloblast-like cells, gland-like structures, and cystic areas. Calcified, odontogenic dentin cement-like deposits were encountered in the stroma of connective tissue in the tumor (Figure 3). With these findings, histopathological diagnosis of AOT was made. After the operation the patient was called intermediate controls (Figure 4). Approximately, seven months after the operation, when bone and tissue healing were nearly complete, the patient moved to another area, and the impacted canine tooth was extracted in another center because of pain in the right upper premolar teeth. Over the course of a three-year follow-up period, stabilization was provided for the teeth that had roots exposed to migration and had showed mobility (Figure 5). No problems concerning healing or recurrence was observed.

DISCUSSION

The case presented here was classified as an extrafollicular subtype of AOT, as it displayed all the general characteristics of AOT, and the lesion was close to but unrelated to the impacted tooth. The intraosseous lesion was located in the maxilla, which is the most common location for extrafollicular types [7].

The asymptomatic nature of AOT is responsible for large lesions [12–14]. In the present study, the lesion grew asymptotically, and pushed the impacted tooth



Figure 1: Intraoral view of the lesion.



Figure 2: Pathologic specimen and extracted supernumerary teeth.

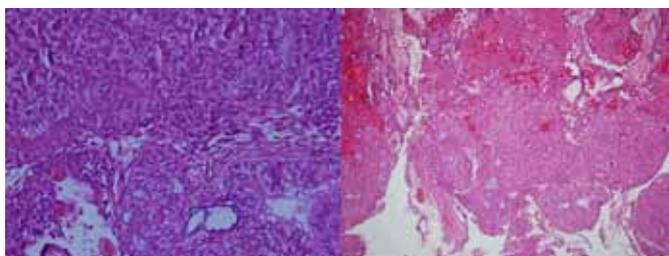


Figure 3: Solid islets, eosinophilic odontogenic hard tissues, glandular structures (H&E stain, x400) and ameloblast-like epithelial cells (H&E stain, x100).



Figure 4: First radiographic view of the region after operation.

far from its normal location, but caused no pain within that period. We believe that the pain experienced by the patient during the healing period before the impacted



Figure 5: Postoperative radiographic view of 3rd year of the lesion region.

tooth extraction originated from continued eruption of the tooth after removal of the suppressing lesion.

The possibility of a radicular cyst was considered in the differential diagnosis of this lesion, which was radiologically unilocular, had definite margins, and included dental roots, but the diagnosis of radicular cyst was eliminated owing to the absence of aspiration material. Root resorption should be considered in AOT even though it is rarely encountered; in the present case, resorption was observed in the root of the central incisor [3, 13]. AOT should primarily be differentiated from calcified odontogenic tumors, as it may contain calcified foci, as was the case in the present study, and from cysts with radiological similarity [3, 14]. In addition, ameloblastoma, ameloblastic fibroma, and ameloblastic fibro-odontoma are lesions that should be considered in the differential diagnosis [15]. AOT is sometimes referred to as adenoameloblastoma owing to the similarities with ameloblastoma and its variants, and conspicuous “preameloblast-like cells” seen on histopathological examination [3].

Terminology for this tumor is debatable and some authors believe that the tumor is an adenomatoid odontogenic cyst [16]. Cases of AOT with concurrent odontogenic cysts have been reported in literature [17, 18]. Histopathological examination of the present case revealed tumor sections consisting of gland-like structures and cystic areas. However, failing to obtain aspiration fluid is incongruous with the term cyst. In our opinion, an adenomatoid odontogenic cyst, which causes confusions in terminology, might be a condition in which AOT is encountered in combination with an odontogenic cyst.

The fact that AOTs are encapsulated provides a therapeutic advantage for surgery, and recurrence has not been reported after conservative surgical treatment of these lesions [19]. In the present study, no recurrence (cystic areas observed on microscopic examination) was observed over the course of more than three years of follow-up, and this supports the notion that conservative surgery is an adequate treatment for such lesions.

CONCLUSION

The present case is important because it describes extrafollicular type Adenomatoid odontogenic tumor (AOT) that caused root resorption, and it is one of the few cases of AOT with long-term follow-up. A long follow-up period is important in cases like AOTs, because when they occur in combination with other lesions or contain a cystic structure the possibility of recurrence must be considered.

Author Contributions

Cennet Neslihan Eroglu – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for intellectual content, Final approval of the version to be published

Serap Keskin Tunc – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Final approval of the version to be published

Omer Gunhan – Substantial contributions to conception and design, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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