

A "RECURRENT" ADENOMATOID ODONTOGENİC TUMOUR CASE REPORT

Fevzi YAVAŞ (*) • Misten DEMİRYONT (**) • Atilla ARINCI (*)

Cengiz ERSEZEN (*) • Sıtkı TUZLALI (**)

SUMMARY: Adenomatoid odontogenic tumour (AOT) is a benign odontogenic tumour and accepted as non-recurrent even after incomplete removal practically. A case of recurrent AOT confirmed with microscopic study is presented and the literature reviewed.

The adenomatoid odontogenic tumour is an uncommon epithelial odontogenic tumour of benign character. It has been described under a variety of different names in the literature, probably because of the controversy regarding to pathogenesis and cell origin (1,6,9,13,15,19). It has been first shown by Dreibladd in 1907 and described by Stafne in 1948 as a special entity.

AOT's treatment is simple enucleation and no recurrence has been reported -even after incomplete removal - in the literature. In this article we present an AOT case which recurred in 3 years.

CASE REPORT

A male patient, 24 years old, presented with a vegetative swelling on the right side of his palate, near the canine tooth. A partial maxillectomy was performed. Macroscopically the tumour, 5 cm. in diameter, presented a dark red capsule. Its cut section revealed a yellow-white appearance with a soft consistency, containing some harder areas. In sectioning, an embedded tooth was recognized (Figure 1).

Microscopic examination of the specimen revealed islands of cuboidal or columnar shaped cells which formed lobule-like structures and spindle cells between them forming strands and surround the solid island (Figure 2). The lobule-like structures usually contained deposits of homogenous, eosinophilic material resembling amyloid, some of which seemed to be calcified.

It was also seen elsewhere between lobule-like structures. A few of the solid islands had duct-like structures in the center (Figure 3). They were lined with columnar cells with their nuclei oriented at their bases. Eosinophilic material in tumoural islands stained positively with PAS stain, but did not stain with Kongo stain. Alcification was seen in lobule-like structures and in the stroma.

Two years later on the same (left) border of resection, another mass of 4 x 3 x 1 cm. is noticed on routine controls and resected. The histological examination of the specimen revealed AOT and evaluated as a recurrence.

DISCUSSION

AOT is an uncommon, distinct tumour of odontogenic origin (1,6,9,13,15,19). Although it has consistent clinical and histopathological features, probably because of the confusion regarding to the pathogenesis and cell origin, it has been described under various designations in the literature, viz: adenoameloblastoma, cystic complex composite odontoma, tumour of enamel organ epithelium, ameloblastic adenomatoid tumour, adeno-

(*) İstanbul Üniversitesi, İstanbul Tıp Fakültesi,
Plastik ve Rekonstrüktif Cerrahi Anabilim Dalı, Çapa, Topkapı-İstanbul

(**) İstanbul Üniversitesi, İstanbul Tıp Fakültesi, Patoloji Anabilim Dalı,
Çapa, Topkapı-İstanbul



Figure 1: Post-operative view of the specimen.

matoid ameloblastoma, epithelial tumour associated with developmental cysts, unusual pleomorphic adenoma-like tumour (2,3,6,7,9,11,13,17,18). As it can be understood from the nomenclature, initially several authors suggested that the AOT was a variant of ameloblastoma which could lead to tragic errors in treatment.

AOT was first shown by Dreibradt in 1907 and described as a distinct tumour by Stafne (15) in 1948. In 1969 Philipsen and Birn were the first to use the term AOT for the tumour (1,6,9,19,20).

AOT, with its clinical behaviour and histological features, is a benign odontogenic tumour. The majority of patients is in the second decade (% 70). Our patient is also 24 years old. Women predominate men with a ratio of % 64. The age ranges between 5 and 48 years (5,12). Its localisation is also characteristic, predominantly on the anterior maxilla, especially in the canine teeth region (1,4,6,8,9,19). This suits with the localisation of the lesion in our case. Mandibular involvement is rare.

It may be asymptomatic or the only clinical sign may be the delayed eruption of teeth and a fair painless swelling in the region (1,4,6,9,16,17,18,19). The patient's complaint was only swelling without pain in our case. Although a few large tumours have been reported, the lesion is between 1.5-3 cm in diameter, it occasionally causes asymmetry of the face. In our case the primary lesion was 7x7x6 cm. Fluctuation and crepitation can be found in cases with a cyst (8,19). Dental row deformities can be present.

Radiographs display a well-demarcated, radiolucent, lytic, sclerotic lesion frequently associated with an embedded tooth (1,4,6,9,19,20). Small foci of calcifications can be featured inside the lesion, and these, together with the presence of an embedded tooth, are pathognomonic for AOT especially in differentiating from a dentigerous cyst (8).

Microscopic examination reveals circular masses composed of columnar to cuboidal epithelial cells. These masses are named as "adenomatoid lobules" by Tahsinoğlu (18). Inside these, small spaces resembling ductule lumens which are lined by a single layer of

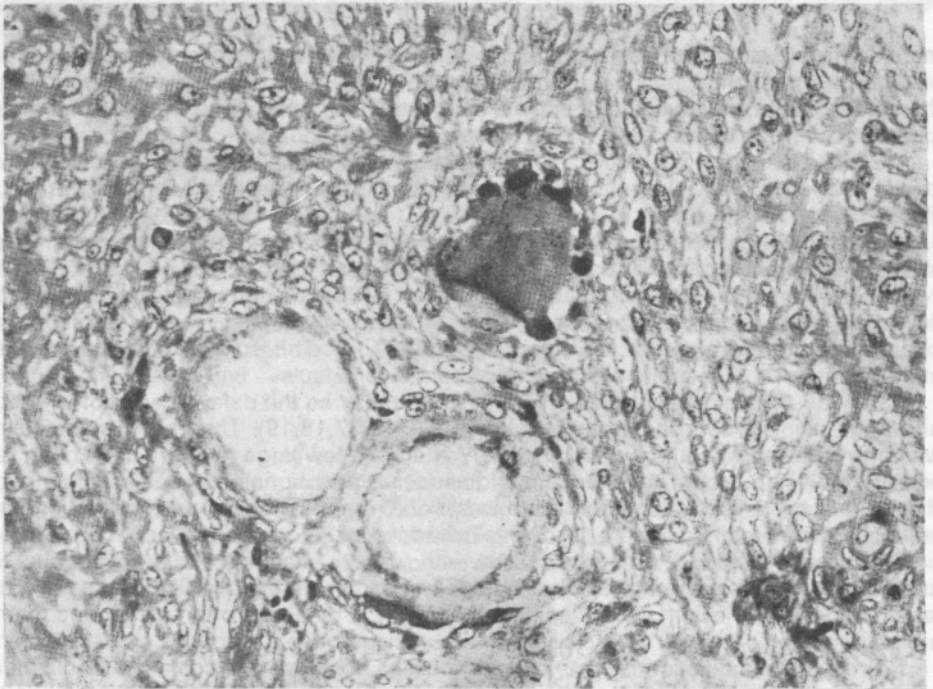
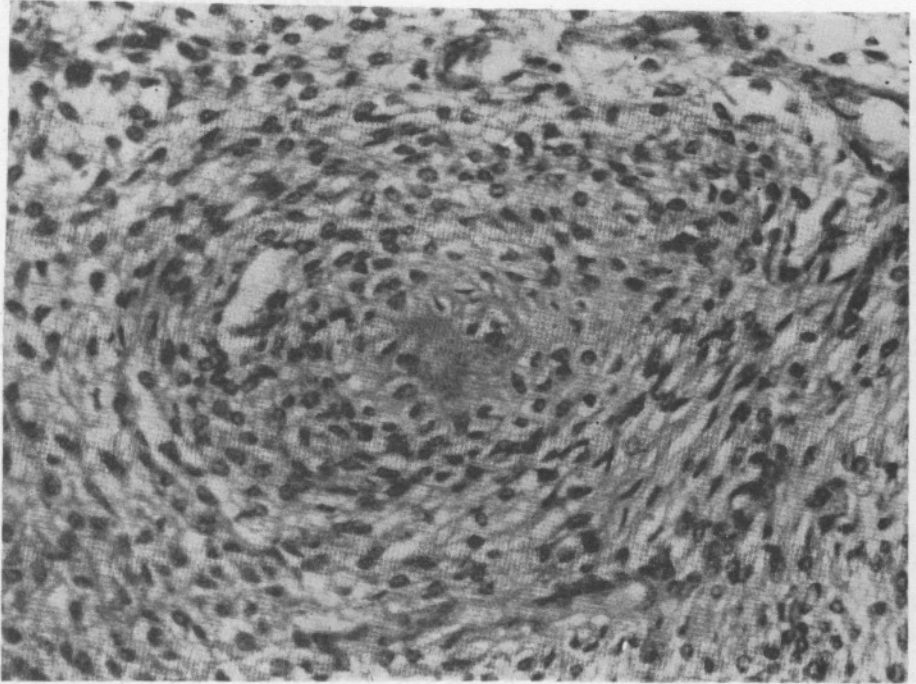


Figure: 2,3. Views of the microscopic sections.

similar cells are seen. There is an eosinophilic amorph substance between the cells forming epithelial nodules and within the lumens of duct-like structures (1,13). The nature of this material has not been well understood yet. According to the study of Smith et al. (13), it resembles amyloid histochemically and ultrastructurally. But our histochemical study did not prove an amyloid origin. Between the epithelial nodules, scattered, sometimes forming strands, there are flattened, spindled cells resembling the stellate reticulum cells and cells of stratum intermedium (10).

The origin of AOT is still controversial. Some authors have speculated that it originates from the wall of a follicular cyst (4,9). Abrams et al (1). considered it as a developmental overgrowth of odontogenic tissue or hamartoma. Spouge suggested the origin from the inner enamel epithelium (14). The detailed ultrastructural works of Smith et al. (13) and Hatakeyama et al. (10) supports the enamel organ origin. These authors observed that the cells of AOT were ultrastructurally similar to the cells of the enamel organ of anormal tooth germ.

Treatment consists of surgical enucleation and no recurrence has been reported even after incomplete removal-its main difference from ameloblastoma (1,6,9,17,18,19, 20). In our opinion although it is very rare, there is a probability of AOT to recur and this must be kept in mind when determining the resection limits.

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