Journal of Oral Science, Vol. 43, No. 4, 283-286, 2001

Case report

An adenomatoid odontogenic tumor with unusual clinical features

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(Received 2 April and accepted 9 October 2001)

Abstract: Adenomatoid odontogenic tumors are uncommon odontogenic lesions characterized by duct-like structures that form from the epithelial component of the lesion. Most of these masses develop in the second or third decade of life, and there is a strong female bias in occurrence. Typically, these lesions arise in the lateral incisor/canine region of the maxilla, where they produce a swelling. Only in very rare cases is the lesion found distal to the premolar area. Nearly all of these growths are associated with an embedded anterior maxillary tooth (usually a canine), and most resemble a 1-3 cm diameter dentigerous cyst. Radiopacity is reported in two-thirds of cases. This article describes the case of a 9-year-old Caucasian male who presented with a painless swelling in the left premolar-molar region of his maxilla. This case is of particular interest because the features (patient age, gender, lesion location, size, and radiographic findings) were not typical of adenomatoid odontogenic tumor. (J. Oral Sci. 43, 283-286, 2001)

Key word: adenomatoid odontogenic tumor.

Intoroduction

Adenomatoid odontogenic tumors are rare growths that originate from odontogenic epithelium. The lesion is generally considered to be developmental rather than neoplastic in origin (1-4). A review by Philipsen et al. noted that 499 cases of adenomatoid odontogenic tumor had been reported in the literature as of 1991 (3). These masses usually arise in the second or third decades of life, and only a few cases have been reported in children younger than 10 years (3,5). There is a marked bias toward females in the development of this lesion, with a frequency of 64% percent in women and 36% in men (1,2,6). The maxilla is more often involved than the mandible, at a reported ratio of almost 2:1 (2,3,6,7). Three quarters of these lesions involve the anterior aspect of the jaws, and the canine region of the maxilla is the most common site of development. Only rarely is the molar area involved. In about 60% of cases, the mass is associated with an embedded tooth, usually a canine (6,7).

Radiographically, the typical adenomatoid odontogenic tumor appears as a unilocular, clearly defined, radiolucent area that is often associated with an embedded tooth (3,5,7). Sometimes calcifications can be seen within the cyst-like radiolucent zone (1-3,6-8). When the lesions are located between anterior teeth, divergence of roots may be seen, but root resorption is rare (3,6,8,9). Although larger lesions have been reported, the typical diameter is 1-3 cm (3,6,7). Very rare extraosseous cases have also been described (3).

Histologically, the mass consists of cells that are considered to be of epithelial origin. These may be arranged in solid clumps, as duct-like structures, in whorls, in rosette-like formations, or as sheets of spindle-type cells. The duct-like structures may be lined with columnar or cuboidal cells. It is not unusual to find strands and islands of this same epithelium in and around the fibrous capsule, and sometimes calcifications are observed among the "ducts" (1-3,6,7,10).

Other lesions that might be included in a differential diagnosis of adenomatoid odontogenic tumor are dentigereous cysts, because of frequent association with impacted teeth and lateral root cysts, because of occasional

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location adjacent to roots of anterior teeth (9). Conservative treatment (enucleation) is all that is required for this lesion, because the adenomatoid odontogenic tumor is a totally benign encapsulated lesion that very rarely recurs.

This report describes an adenomatoid odontogenic tumor that was highly unusual with respect to the age and gender of the patient, and the site and extent of the lesion.

Case Report

A 9-year-old Caucasian boy was referred to the Department of Oral Surgery at the Hacettepe University Faculty of Dentistry with the complaint of a swelling in the left upper jaw. The patient had a slow-growing, painless, gingival swelling in his left maxillary premolar-molar region for the past 3 years. There was no history or any signs of systemic disease.

At the time of examination, the boy's face was asymmetrical due to generalized swelling of the left maxilla. There was no regional lymphadenopathy. An intraoral examination revealed a well-defined, 3×4 cm, granulomatous mass in the left posterior region of maxilla. Although the lesion had perforated the buccal plate, palpation did not induce bleeding.

Radiographic and computerized tomographic evaluations demonstrated a mixed radiolucent-radiopaque lesion with a well-defined border that extended from the right central incisor to the left first-molar region (Fig. 1). The x-rays showed a unilocular, clearly defined, radiolucent lesion associated with the first premolar. Displacement of the premolars and separation of the roots of the incisors were also evident. In addition, we noted an irregular form of root resorption on the distal surface of the left central incisor.

An incisional biopsy was obtained under local anesthesia. The histological findings in the biopsy and subsequent total excision specimen were essentially similar. The pathological diagnosis was adenomatoid odontogenic tumor with no

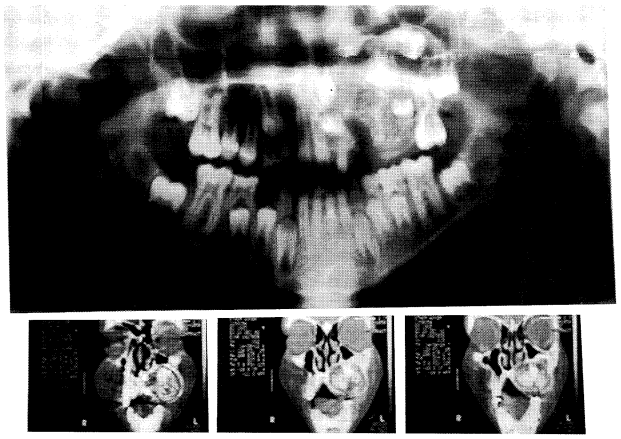

Fig. 1 Preoperative panoramic radiography and computerized tomography views of the affected site.

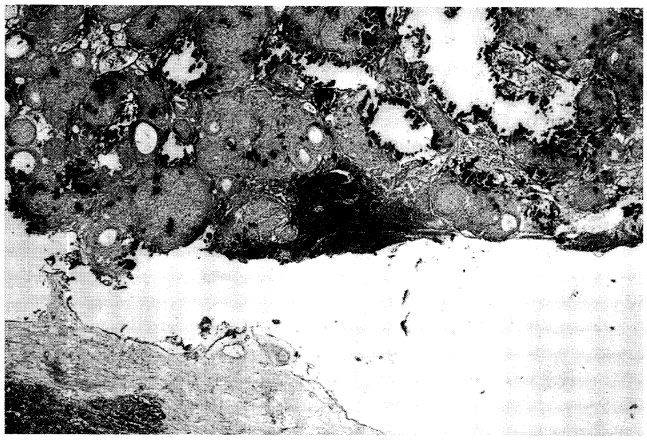

Fig. 2 Section from the wall of the cystic cavity and the neoplastic tissue. The cystic space was lined by flattened, single-layered epithelium in this area. (HE ×115)

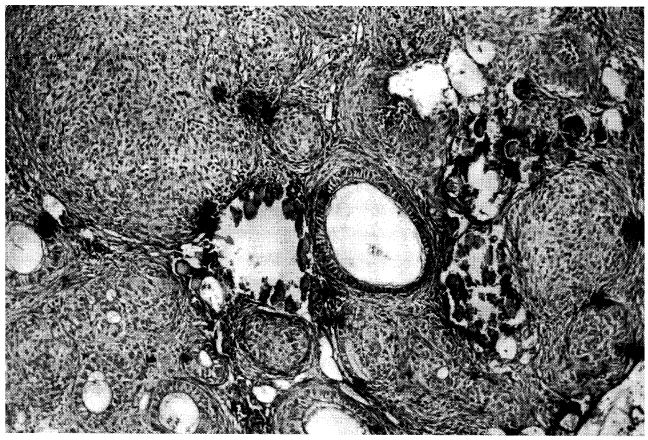

Fig. 3 Higher magnification of the neoplasm showing ductal structures and whorls of epithelial tissue, cellular stroma and foci of calcification. (HE ×230)

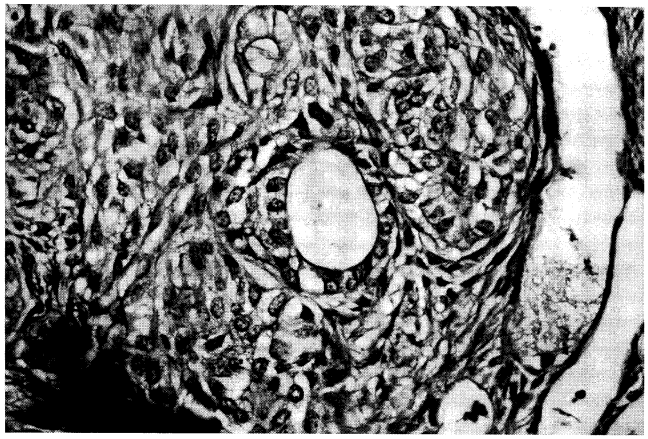

Fig. 4 Transition between the columnar cells lining the ductal structures and spindle cells of the stroma. (HE ×460)

evidence of malignancy. The excision material consisted of irregular fragments of pink soft tissue with foci of calcified material.

Surgery was performed with the patient under general anesthesia. The tumor was encapsulated and approximately 5 cm in diameter, and we were able to totally excise it from its crypt. In addition, we extracted all the embedded teeth in the affected region of the maxilla. Only the second molar was left intact, as it was not associated with the lesion. The postoperative course was uneventful.

Histological examination of the incisional, as well as excisional, biopsy specimens showed neoplastic tissue located within a cystic cavity lined by single or stratified epithelium (Fig. 2). The neoplasm revealed ductal formations lined by columnar epithelial cells, whorls of spindle or polygonal cells and a cellular stroma composed of spindle cells (Fig. 3). In some areas, transitions between the cells lining the ductal formations and stromal cells were noted (Fig. 4). Irregular deposits of calcified material were observed mostly in the stroma. There was no evidence of cellular atypia and no increase in mitotic activity was evident. These findings were fully compatible with the diagnosis of adenomatoid odontogenic tumor.

Discussion

Gorlin et al. classified the adenomatoid odontogenic tumor as a neoplasm of odontogenic epithelium that has no inductive effect on the connective tissue (1); however, several cases have suggested that this classification was premature (1-3). Some authors believe that the lesion is a developmental outgrowth or hamartoma, whereas others consider it a neoplastic growth of odontogenic epithelium (3). It was even known by the name of adenoameloblastoma; clinically, histologically, and behaviorally it is clearly different from ameloblastoma (9). The 1971 World Health Organization classification states, "It is generally believed that the lesion is not a neoplasm."

Philipsen et al. (3) categorized adenomatoid odontogenic tumors as three variants based on clinical and radiological findings. These variant forms are a central follicular type, a central extrafollicular type, and a peripheral variant. Clinically, all these forms are characterized by slow but progressive growth accompanied by few or no symptoms (1,3,6). Expansion of the cortical bone is a common finding in the central variants, but penetration of the cortical plate is unusual (3). Especially in the extrafollicular type, there may be some displacement of neighboring teeth, but root resorption is rare, and the size of central lesions generally varies from 1-3 cm diameter (3,6-8). Radiographs show a well-demarcated area of radiolucency, and central lesions may also contain fine, sharply defined, radiopaque foci of calcified tissue (3,7).

The presented case of adenomatoid odontogenic tumor is of particular interest because it was unusual in terms of the patient's age and gender, the lesion's location and size, and the radiographic findings. The affected individual was a 9-year-old male and the mass was located in the posterior part of his maxilla. The lesion was unusually large and had perforated the buccal plate. Based on the Philipsen classification, the presented case belonged to the follicular type of central variant extended to soft tissue. There was also cystic radiolucency, but the embedded tooth was the first premolar as opposed to the more typical canine. As another unusual feature, we noted irregular root resorption on the distal surface of the left central incisor. The treatment for this case involved enucleation and curettage. The patient has been re-checked every 3 months for 3 years, and there has been no recurrence to date.

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