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CASE REPORT

Adenomatoid odontogenic tumor: a report of a rare case with recurrence

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Adenomatoid odontogenic tumor (AOT) is an uncommon tumor of odontogenic origin, composed of odontogenic epithelium in a variety of histoarchitectural patterns, and characterized by slow but progressive growth. Generally, recurrences seldom, if ever, occur after surgical curettage, while here we report an extremely rare case of AOT with recurrence. The patient was a 36-year-old Chinese man who presented with a palpable bony-hard swelling in the anterior maxillary region initially at the age of 16 and the lesion recurred twice over a 20-year period. The tumor was diagnosed as AOT with well-defined unilocular radiolucency shown in X-ray graphs and solid nodule of cuboidal or columnar cells of odontogenic epithelium forming typical nests or rosette-like structures as well as characteristic duct-like spaces in histologic findings.
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Case report

In September of 1986, a 16-year-old boy presented with swelling in the upper right canine region. Examination revealed a well-defined, unilocular radiolucent lesion associated with an unerupted upper right canine. It was enucleated and a diagnosis of dentigerous cyst was made clinically. There was no biopsy for confirming the disease.

In November of 1998, the patient, now 28 years old, was admitted to Peking University School and Hospital of Stomatology with a right upper jaw swelling with tooth resorption of 1-year duration. Examination revealed that a palpable bony-hard swelling had ill-defined margins with a surgical scar on the overlying mucosa, which implied the lesion arised again in the same region as that found 12 years ago. A well-defined, unilocular radiolucency was shown on X-ray involving the teeth from upper right incisor to the maxillary right second premolar (Fig. 1). The possibility of apical cyst was considered clinically. After surgical curettage with extraction of the maxillary right first and second premolars, gross examination of the specimen showed a single well-encapsulated grey-brown soft tissue measuring $4 \times 3 \times 2.5$ cm (Fig. 2). Microcopy revealed conspicuous nests or rosette-like structures as well as tubular or duct-like appearance formed by cuboidal or columnar cells of odontogenic epithelium, which confirmed the diagnosis of adenomatoid odontogenic tumor (AOT; Fig. 5a). Staging workup did not detect any other evidence of the disease. The patient was followed without treatment.

In March of 2006, the patient, now 36 years old, was referred to the same surgeon with a palpable bony-hard swelling in the anterior maxillary right region of several months duration. It was excised in toto and the enucleated tissue was reported as recurrent AOT. Radiographically, the lesion showed a well-defined, unilocular radiolucency involving the teeth from upper right incisor to the maxillary right first molar. Macroscopically, the lesion was well circumscribed and a single soft mass, measuring $4.5 \times 4 \times 2.5$ cm, and cut surface showing a gray cystic appearance (Figs 3 and 4). Microscopically, the relapsed specimen showed similar histologic findings to the initial one, presenting as cuboidal or spindle-shaped epithelial cells forming aggregates or typical rosette-like structures with minimal connective tissue, and cuboidal or low columnar cells forming glandular duct-like structures (Fig. 5b,d). Between the epithelial cells, eosinophilic amorphous material (tumor droplets) and globular masses of calcified substances were also found (Fig. 5c). The patient is under follow up after local excision.

Comments

Adenomatoid odontogenic tumor is an uncommon tumor of odontogenic origin, defined by WHO as a

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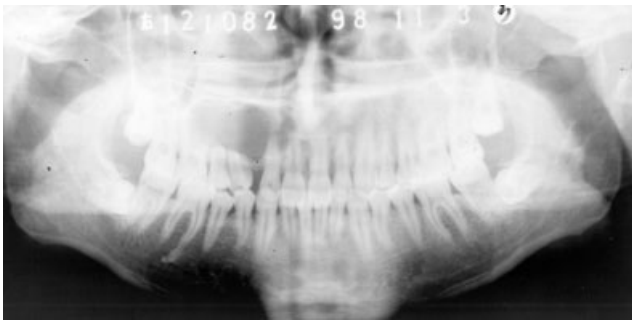

Figure 1 Panoramic radiograph showing an unilocular well-circumscribed radiolucency extending from upper right incisor to the maxillary right first molar, with resorption of root apices of the maxillary right two premolars and the displacement of the maxillary right first molar.

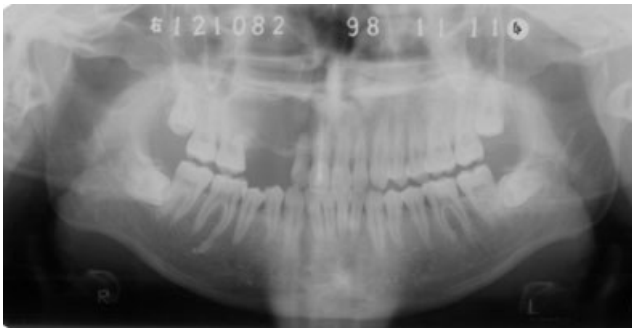

Figure 2 Panoramic radiograph showing an unilocular well-circumscribed radiolucency extending from upper right incisor to the maxillary right first molar, with the maxillary right premolars missing and the displacement of the maxillary right first molar.

Figure 3 Gross-examination of the specimen showed a single well-encapsulated grey-brown soft tissue measuring 4 × 3 × 2.5 cm.

tumor composed of odontogenic epithelium in a variety of histoarchitectural patterns, embedded in a mature connective tissue stroma and characterized by slow but progressive growth (1). Sometimes AOTs are also considered as a hamartoma with completely benign behavior that can be cured by local excision

Figure 4 The cut surface of the tumor showed a cystic appearance.

(2). Most arise in females during the second or third decade of life, often in the anterior maxilla. Generally, recurrences seldom, if ever, occur after surgical curettage, while we are reporting here an extremely rare case that recurred twice over a 20-year period and; however, it was really typical in clinicopathologic characteristics, presenting as a well-circumscribed intraosseous lesion in the anterior maxillary right region with slow but progressive growth. At low magnification the most striking histoarchitectural pattern was that of variably sized solid nodules of cuboidal or columnar cells of odontogenic epithelium forming nests or rosette-like structures with minimal stromal connective tissue. Between the epithelial cells, eosinophilic amorphous material (tumor droplets) and globular masses of calcified substances were also present. Conspicuous within the cellular areas were structures of tubular or duct-like appearance. In addition to forming ducts, the cuboidal to columnar cells also formed convoluted cords in complicated patterns that exhibit invaginations. All these above clinicopathologic features found in the case reported here confirmed the diagnosis of AOT.

There are three variants for AOTs, named as follicular, extrafollicular, and peripheral, and all variants show identical benign biologic behavior and almost all are encapsulated. The more common variant is the follicular type, which involves an unerupted tooth and is often mistaken for a dentigerous cyst (3). In this case, the initial lesion found in 1986 presented as a well-defined, unilocular radiolucency associated with an unerupted upper right canine, which was then diagnosed as dentigerous cyst clinically without any biopsy for confirming the disease. As the intraosseous follicular AOT, as this case reported here, frequently resemble dentigerous cysts, and the cystic mass found in 1986, 1998, and 2006 arised in the same region with the latter two cystic masses confirmed as the diagnosis of AOT by histopathology, the diagnosis for the cystic mass containing an unerupted upper right canine in 1986 would rather be referred as AOT not dentigerous cyst, and then

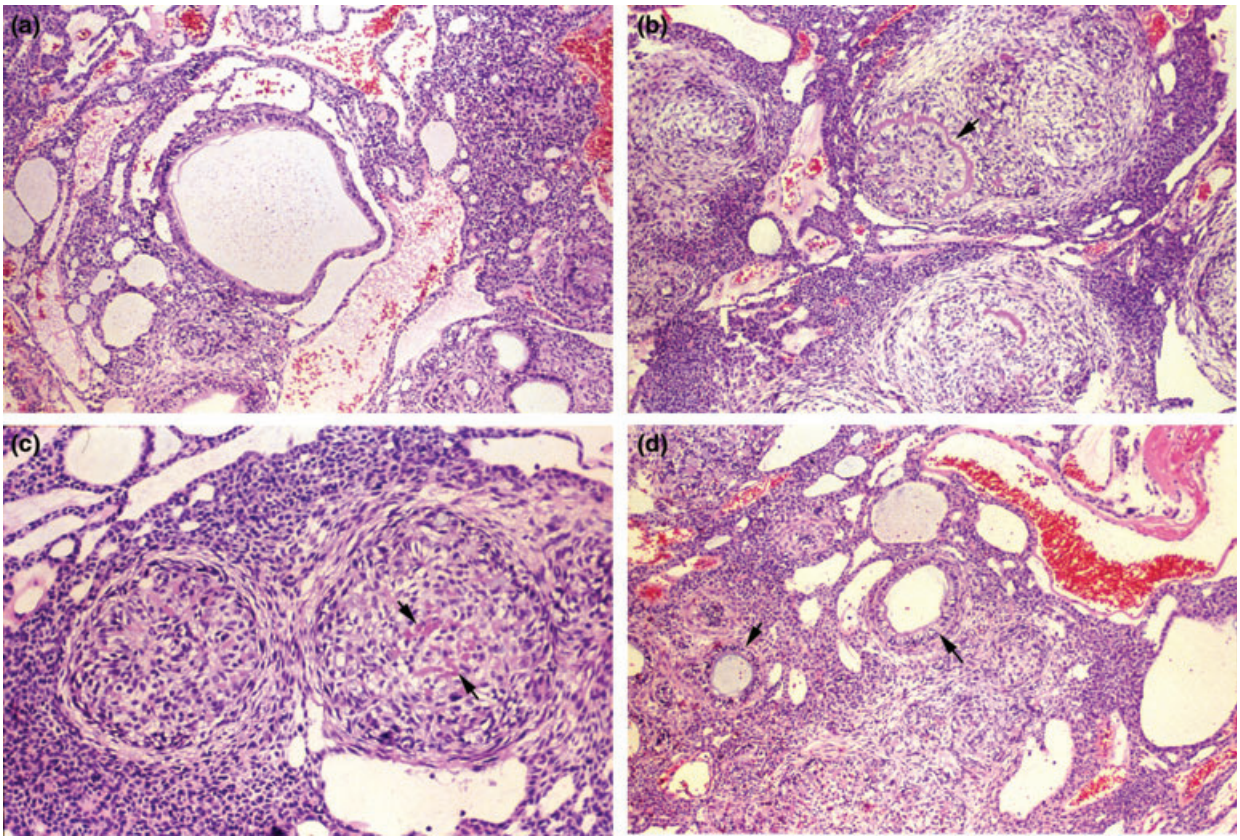

Figure 5 Histopathologic findings. (a) Photomicrograph in 1998 showed features typical of adenomatoid odontogenic tumor with cuboidal or columnar cells of odontogenic epithelium forming duct-like structures and nests [hematoxylin and eosin (H & E), original magnification ×75]. (b) Photomicrograph in 2006 showed solid nodules or duck-like structures, together with a rosette-like structure (arrow; H & E, original magnification ×75). (c) Photomicrograph in 2006 showed solid nodules of cuboided epithelium containing several eosinophilic, amorphous 'tumor droplets' (arrows; H & E, original magnification ×150). (d) Photomicrograph in 2006 showed duct-like structures lined by a single row of columnar epithelial cells (arrows; H & E, original magnification ×75).

the lesion was considered to have recurred twice over a span of 20 years.

Interestingly, our present case has some unusual clinical findings from most normal types of AOT. It is reported that the intraosseous AOTs may cause displacement of neibouring teeth, but root resorption is a very less common finding (3). In fact, to the best of our knowledge, only three cases of AOT with root resorption have been reported (4–6); however, the case reported here presented with resorption of root apices of the maxillary right two premolars and the displacement of the maxillary right first molar. In addition, most AOTs are asymptomatic and usually do not exceed 1–3 cm in great diameter (3, 5), but larger lesions have been reported in the literature (6), as the case reported here presenting as a palpable bony-hard swelling measuring 4 × 3 × 2.5 cm with pain. Meantime, it is reported that the female:male ratio is 1.9:1 (3) and even higher ratios are found in Asian populations, the highest incidence being observed in Sri Lanka (3.2:1; 7) and Japan (3:1; 8). While there is this strong female bias in occurrence, AOTs are also found in male patients, as in the case reported here the patient was a 36-year-old Chinese man.

Furthermore, to our knowledge, AOTs are widely recognized as to be cured by local excision with extremely rare recurrences. According to the review study of AOT biologic profile based on 499 cases by Philipsen et al. (3), conservative surgical excision is the treatment of choice and documented recurrences had not been reported till 1991 (9). In addition, Handschel et al. reviewed all reports regarding AOT and cited in 'PubMed' since 1990 and found the prognosis for AOTs is excellent with conservative treatment, such as curettage or enucleation (10). In fact, as far as we know, only three cases of recurrent AOT among more than 750 cases have been reported and all the recurrent cases were in the Japanese literature (8, 9, 11). However, here in our present case, with conservative treatment by enucleation in the first time in 1986, the tumor recurred 12 years later in 1998, and after conservative treatment by currettage in 1998 the tumor recurred again in 2006. As AOTs have always been considered as a tumor with benign behavior that can be cured by conservative treatment, this present study may provide a refresher for general dental practitioners about prognosis and predictive factors of AOTs, and careful follow-up examinations should be conducted in this disease.

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