CASE REPORT

Adenomatoid odontogenic tumor originated in the periodontal ligament

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Summary Adenomatoid odontogenic tumor (AOT) is a benign (hamartomatous), non-invasive lesion with slow but progressive growth. There are three variants of AOT: follicular, extrafollicular, and peripheral. This report illustrates an unusual case of AOT, i.e., arising within the periodontal ligament. A boy was brought in by his parents for evaluation of a gingival swelling. Periapical radiograph revealed thickening of the periodontal ligament with foci of calcification. Clinical diagnosis was peripheral ossifying fibroma. Biopsy was performed and microscopic examination revealed the presence of an AOT. The patient has been followed-up for eleven months without recurrence.

Introduction

Adenomatoid odontogenic tumor (AOT) is a benign (hamartomatous), non-invasive lesion with slow but progressive growth that accounts for 2.2–13% of all odontogenic tumors. AOT usually affects young patients, mostly during their second decade of life. There is a tendency to affect females (male: female ratio 1:2) and to occur in the anterior maxillary region.1–4

There are three variants of AOT: follicular, extrafollicular, and peripheral. The follicular type is a central intrabony lesion associated with an unerupted tooth, which accounts for about 70% of all cases. The extrafollicular type is also an intra-ossuous lesion, but unrelated to an unerupted tooth, and represents 25% of all AOTs. The peripheral type is a rare form that arises in the gingival tissue, and only 18 well-documented cases were reported.2,3 All three variants have the same histological aspect and clinical behavior.5

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Figure 1  (A) Clinical aspect showing a firm, sessile, predominantly normally-colored, smoothly surfaced, 2 cm swelling in the anterior right maxillary gingiva. Also, there was a focal and well-demarcated red-colored area in the region adjacent to the marginal gingiva. (B) Periapical radiograph revealed thickening of the periodontal ligament (arrow heads) and foci of calcification (arrow). (C) Expansion of the buccal osseous cortical in the region of tooth number 12 and lesion found to be located within the periodontal ligament during surgery. (D) Surgical removal using a curette.

Figure 2  (A) Sheets and strands of odontogenic epithelium (haematoxylin–eosin stain; 200× magnification). (B) Strands of odontogenic epithelial cells composed of predominantly cuboidal and occasionally spindle-shaped cells. Also, eosinophilic amorphous mineralization (arrow) was seen (haematoxylin–eosin stain; 400× magnification). (C) Eosinophilic amorphous mineralization (arrow) and duct-like arrangement were present (arrow heads) (haematoxylin–eosin stain; 400× magnification).
This study aimed to describe an unusual case of AOT, originating in the periodontal ligament region of a 15-year-old Brazilian boy.

Case report

A 15-year-old Caucasian boy was brought in by his parents on May 2005 to the Stomatology Clinic of the Universidade Federal de Minas Gerais (UFMG) complaining of a 9-month asymptomatic gingival swelling. The boy was undergoing orthodontic treatment over three years. Intra-oral physical examination revealed a firm, sessile, predominantly normally-colored, smoothly surfaced, 2 cm swelling in the anterior right maxillary gingiva. Also, there was a focal and well-demarcated red-colored area in the region adjacent to the marginal gingiva (Fig. 1A). Periapical radiograph showed widening of the periodontal ligament with foci of calcification. The lamina dura was well preserved (Fig. 1B). Clinical diagnosis was peripheral ossifying fibroma. The patient was submitted to an excisional biopsy. During flap reflection, expansion of the buccal osseous cortical was observed and the lesion was found to be located in the buccal periodontal ligament of tooth #12 (Fig. 1C and D). Microscopic examination revealed sheets and strands of odontogenic epithelium composed of predominantly cuboidal and occasionally spindle-shaped cells. Scattered duct-like structures were observed as well as occasional droplets of calcification and small foci of acellular, eosinophilic amiloid-like material (Fig. 2A–C). Final diagnosis was AOT. The patient has been followed-up over eleven months without recurrence (Fig. 3A and B).

Discussion

Clinical, radiographic, and trans-surgical features of the reported AOT case show that it was located within the periodontal ligament. AOT has three different clinic-topographic forms: follicular, extrafollicular and peripheral. According to Philipsen et al., extrafollicular AOT presents itself as a radiographic image (radiolucency with or without radiopaque points) that may be found in four different locations: above, between, superimposed at midroot level, or superimposed at apex level of erupted teeth. The current case was an extrafollicular AOT without the classic radiographic aspect, causing bone expansion rather than resorption. In addition, our case was clinically similar to a peripheral AOT, i.e., a fibroma or epulis attached to the labial gingiva. Therefore, the reported case showed features of both intra-osseous and peripheral forms of AOT.

Conservative surgical enucleation or curettage has proven to be the treatment of choice for all types of AOTs. All forms show identical benign biological behavior, and in only three out of 750 cases, recurrence of this tumor was verified. In the case reported, the lesion was surgically enucleated, with no signs of recurrence eleven months after surgery.

AOT is an odontogenic tumor whose origin is still controversial. Some but not all of the follicular types of AOT may derive from the odontogenic epithelium of a dentigerous cyst. Dental lamina remnants likely represent progenitor cells for the peripheral type of this benign odontogenic tumor. Following entrapment, these epithelial remnants proliferate in response to an unknown stimulus, giving rise to the lesion. Furthermore, Malassez remnants found in the periodontal ligament may possibly give origin to an extrafollicular AOT, such as in the reported case.

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References


