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CASE REPORT

Adenomatoid odontogenic tumor: an unusual histological presentation with "Pindborg-like" differentiation

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Abstract:

Adenomatoid odontogenic tumor (AOT) is a relatively rare odontogenic tumor derived from odontogenic epithelium. Although there have been many reports regarding the histological spectrum of AOT, only a few have highlighted unusual histological presentations. A 28-year-old female presented with a painless swelling in the maxillary anterior region. Radiological examinations revealed a well-defined unilocular lesion, with sclerotic margins, containing foci of slightly radiopaque material and promoting root divergence between teeth 22 and 23. An incisional biopsy showed spindle-shaped cells forming sheets, strands, and whorled masses, together with ductlike structures, throughout a scanty fibrous stroma. In some areas, sheets of polyhedral epithelial cells with prominent intercellular bridges, mild pleomorphism, and droplets of amyloidlike material were observed. The diagnosis was AOT with "Pindborg-like" differentiation. The occurrence of such differentiation in AOT seems to represent the caricatures of the enamel organ itself and is believed to be an altered phenotype of the tumor.

Keywords: Odontogenic Tumors; Maxilla; Neoplasms.

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INTRODUCTION

The latest WHO classification of odontogenic tumors defines AOT as being a proliferation of odontogenic epithelium exhibiting a variety of histoarchitectural patterns embedded in mature connective tissue stroma characterized by slow and non-invasive growth[1]. On the other hand, calcifying epithelial odontogenic tumor (CEOT) has been defined as a locally invasive epithelial neoplasm with a well-recognised histopathological pattern, characterized histologically by the presence of polygonal epithelial cells, calcification, and eosinophilic deposits resembling amyloid[2,3]. Compared to CEOT, AOT is a non-aggressive tumor, which exhibits limited growth and accounts for about 3% to 7% of all odontogenic tumors[4].

Although the presence of intratumoral nodules of epithelial cells similar to the CEOT has been reported in AOTs[5-10], and some authors have considered these areas as true foci of CEOT[5,6,11-14], it is important to emphasize that the cases reported previously[5-14], as well as presented here, have behaved as typical AOT cases, with no recurrence or aggressiveness. However, it is important to emphasize that these findings may present difficulties for the diagnosis or choice of appropriate therapy. Thus, the purpose of this work is to report a uncommon case of AOT with "Pindborg-like" differentiation and discuss the significance of this histopathological finding.

CASE REPORT

A 28-year-old female patient, caucasian, sought the service of the stomatology clinic at Tiradentes University (Aracaju/SE) complaining of swelling in the maxilla. The intraoral examination revealed a painless volume increase in the anterior region of the maxilla, extending from tooth 22 to 24, firm to palpation, covered by normal-colored mucosa and presenting discrete ulcerated areas (Figure 1). Anamnesis revelated evolution time of 5 months. Previous medical history was not contributory. Imaging exams revealed a well-defined unilocular lesion, with sclerotic margins, containing foci of slightly radiopaque material and promoting root divergence between teeth 22 and 23, as well as expansion of the buccal and lingual cortical plates (Figure 2). The provisional diagnosis of AOT was established and an incisional biopsy of the lesion was performed. The surgical specimen (Figure 3) was submitted to the Service of Oral Pathology of the School of Dentistry at Tiradentes University. Histopathological examination of the incisional biopsy revealed cubic and fusiform cells

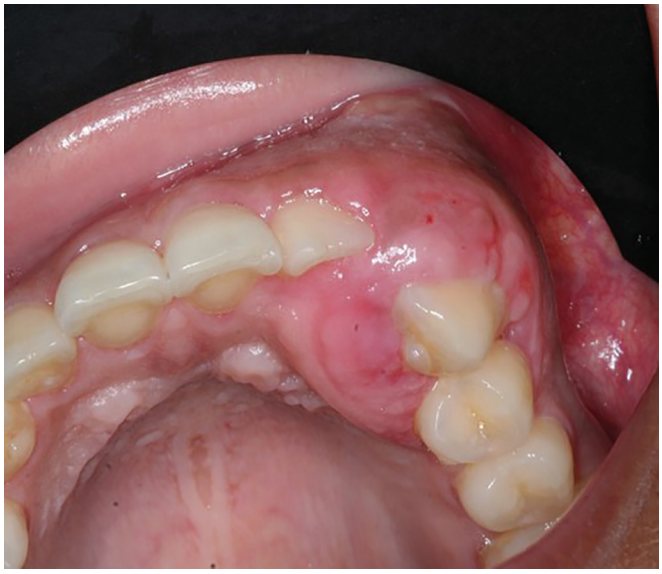

Figure 1. Intraoral aspect of the lesion promoting expansion of the vestibular and palatine cortices.

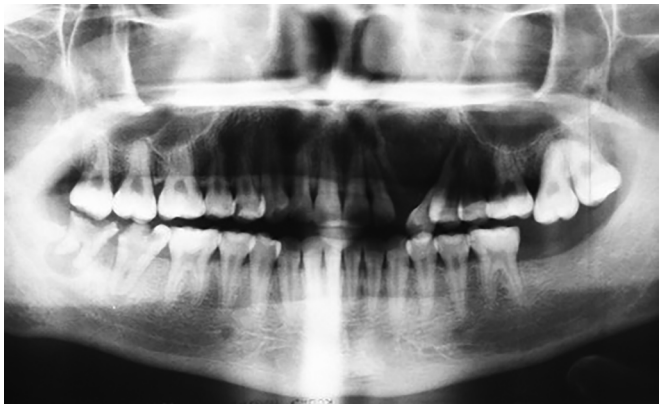

Figure 2. Panoramic radiograph showing unilocular radiolucent lesion with regular margins and well-defined borders promoting root divergence between teeth 22 and 23.

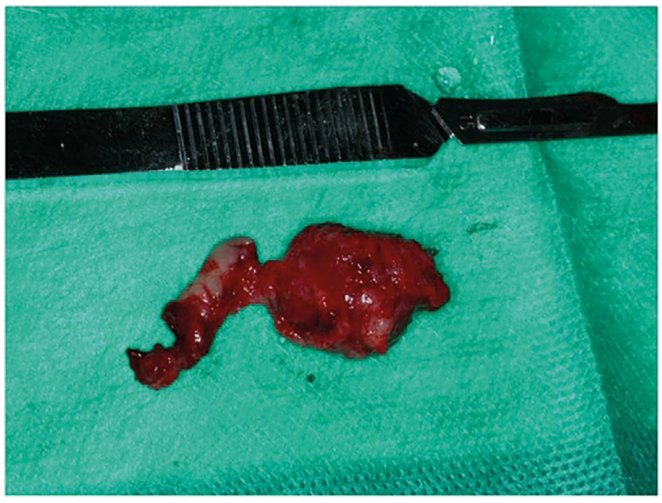

Figure 3. Macroscopic aspect of the surgical specimen removed after enucleation of the lesion.

arranged as nests and rosettes, pseudoductal structures and solid vortex areas. Foci of dystrophic calcification and deposition of dentinoid material were evidente. In some areas, sheets of polyhedral epithelial cells with prominent intercellular bridges, mild pleomorphism, and droplets of amyloid-like material were observed. The diagnosis was AOT with "Pindborg-like" differentiation (Figure 4). The patient was submitted to surgical enucleation of the lesion and is under proservation without signs of recurrence 5 years after surgery.

DISCUSSION

The AOT has three distinct clinicopathological variants: folicular (70%), extrafollicular (25%) and peripheral (about 5% of cases). Radiographically, the follicular variant is characterized as a well delimited radiolucent area, frequently exhibiting discrete radiopaque foci inside and associated with an unruptured tooth, usually the upper canine. The extrafollicular variant is also a central lesion, however, it is not associated with an unruptured tooth and, normally, the radiolucent image is located between two roots, resembling a lateral periodontal cyst[15], as in the case we presented here. Extraosseous or peripheral lesions are very rare and clinically appear as nodules in the maxillary gingiva, mimetizing reactive fibrous lesions of the gingiva[4] and rarely exhibit radiographic imaging; however, slight erosion of the alveolar bone cortex underlying the lesion has been reported[9]. It is important to emphasize that all variants of the AOT share the same histopathological characteristics. In our case, the tumor was an extrafollicular intraosseous type, and also found in the anterior region of the maxilla.

Due to the variety of clinical/radiographic appearances, AOT has several differential diagnoses, varying according to the presence and degree of radiopacity found in lesions and whether or not it is associated with an unruptured tooth. Radiolucent images may mimetize the appearance of dentigerous cysts,

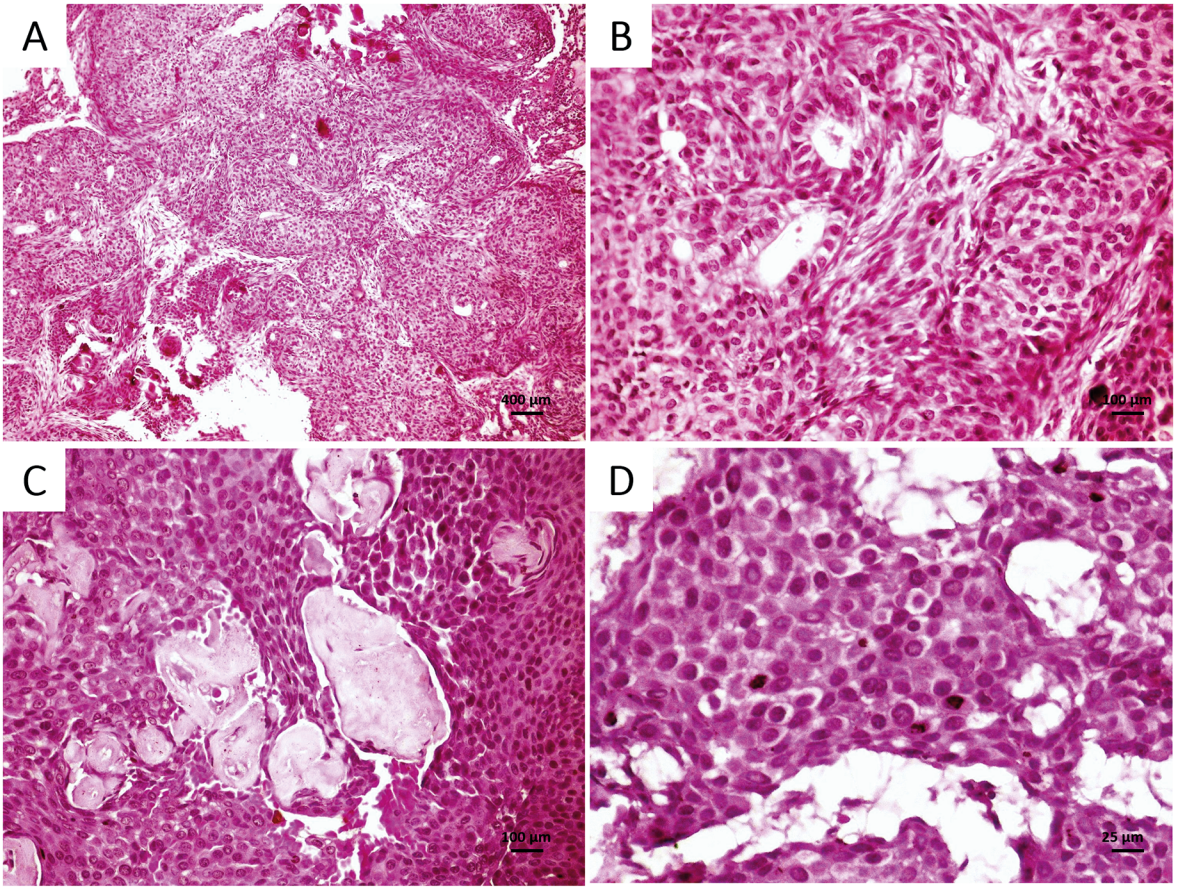

Figure 4. Histological sections stained with hematoxylin-eosin. (A) Compact proliferation of odontogenic epithelial cells forming nodules of cuboid and ovoid cells surrounded by fusiform cells arranged in a vortex pattern (100 x), and (B) forming multiple pseudoductal structures. (C) Sheets of polyhedral epithelial cells with prominent intercellular bridges, mild pleomorphism, and droplets of amyloid-like material (400 x). (D) Detail of polygonal cells exhibiting hyperchromatism and nuclear pleomorphism (800 x).

odontogenic keratocysts, ameloblastomas and lateral periodontal cyst, as lesions exhibiting radiopaque foci can be confused with calcifying cystic odontogenic tumor and even calcifying epithelial odontogenic tumor[4,16].

The epidemiological profile shows predilection for female and young patients, with a peak prevalence in the second decade of life. Clinically, AOT usually manifests as an asymptomatic increase in volume in the anterior region of the gnathic bones, most commonly in the region of upper lateral incisors and upper canines[16]. Thus, this report is in consonance with the clinical characteristics classically described for AOT.

The diagnosis of AOT seldom cause difficulties due to classic histopathological presentations. Histologically, AOT is characterized by the compact proliferation of epithelial cells exhibiting cuboid and/or fusiform shape, permeated by scarce connective tissue stroma. Epithelial tumor cells form sheets, nests, cords, rosette-like structures and pseudoductal structures, formed by cuboidal or columnar cells exhibiting nuclei with polarization opposite to the lumen. Dystrophic calcification foci and areas of deposits of amorphous eosinophilic material with dentinoid appearance are also noted. At the periphery, thick capsule of dense fibrous connective tissue can also be seen surrounding the tumor[1,4,15,16]. CEOT is typically composed by variable amounts of epithelial, amyloid, and calcifying components. The epithelial cells are arranged in sheets, nest and masses of polyhedral cells with abundant eosinophilic cytoplasm, pleomorphic nuclei and prominent intercellular bridges[2,3]. Similar characteristics were observed in the present case.

Although the clinical and pathological features make the diagnosis of the AOT relatively simple, the epithelial component can undergo a series of histological alterations that may hinder the morphological diagnosis, such as the presence of pigmented cells[17], formation of cribriform architectural patterns[18] and differentiation in clear cells[19]. In addition, the AOT may exhibit foci of polygonal cells, with discrete hyperchromatic and pleomorphic nuclei, and formation of amyloid deposits, recalling the findings observed in the CEOT (Pindborg's Tumor)[5-14], as in the case we presented here. Mosqueda-Taylor et al.[9] emphasized that CEOT-like areas in AOTs do not present as solid, infiltrative nests, as it appears in true CEOT.

Moreover, the CEOT-like areas in AOTs lack the typical pleomorphism that is found in the epithelial component of CEOT and their presence do not influence the biologic behavior and growth potential of AOTs. Thus, it is important to emphasize that these findings constitute merely degenerative changes and it should not be interpreted as a characteristic of aggressiveness. This conclusion is supported by the fact that most of the previously reported cases[5-14], including ours, present clinical, radiographic findings and biological behavior basically identical to those described for classic AOT.

In addition there seems to be no reported cases of combined epithelial odontogenic tumour in which CEOT predominates over AOT, which may possibly prove to be a true combination of two different odontogenic tumours[9]. However, it is important to note that true associations between AOT and other odontogenic lesions have been reported, such as dentigerous cyst[20], odontoma[21], ameloblastoma[22] and odontogenic keratocystic[23]. These associations are not considered as mere degenerative changes, but as true hybrid lesions and, sometimes, with more aggressive biological behavior.

Treatment of CEOT consists of surgical removal, which includes a marginal portion of apparently healthy bone. A minimum 5-year observation period is suggested. Maxillary CEOT cases require more aggressive surgery, since these tumors tend to grow more rapidly and are not circumscribed[24]. However, conservative surgical excision is the treatment of choice for AOTs[4,15,16].

The evident clinical aggressiveness of CEOT[2] compared to AOT, which exhibits limited growth and almost zero recurrence tendency[4,15], makes the precise distinction between these two entities a particularly important issue, which can greatly influence the treatment and prognosis of the lesion. Due to the recurrence of AOT to be exceptionally rare, conservative treatment was safely proposed. The patient underwent surgical enucleation of the lesion and after 5 years of follow-up, no relapse was observed.

CONCLUSION

In conclusion, the clinic-radiographic profile of AOT observed in this study agrees with that commonly reported in the literature. The existence of areas composed of polyhedral eosinophilic cells similar in appearance to CEOT in AOT should be considered normal features within the histomorphologic spectrum of AOT and their presence do not influence the biologic behavior and growth potential of AOTs. Instead, it seems to represent a infrequent histomorphological pattern of AOT and it should not be interpreted as a characteristic of aggressiveness.

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