


Follicular adenomatoid odontogenic tumor of a pediatric patient: a case report

Ana Carolina Lemos
Pimentel ¹

Tila Fortuna ²

Alena Ribeiro Alves Peixoto
Medrado ^{3*} 

Sílvia Regina de Almeida
Reis ⁴

Antônio Márcio Teixeira
Marchionni ⁵

Abstract:

Adenomatoid odontogenic tumor (AOT) comprises about 3% of all odontogenic tumors. Rare, benign, slow-growing tumor with increase in volume, associated with a lack of tooth eruption, with female predilection. This study aims to report a case of pediatric patient diagnosed with follicular AOT, treated satisfactorily by surgical excision. 11-year-old girl had increase in volume at right maxilla, with asymptomatic evolution of 1 year. Intra-oral examination showed expansion of buccal cortical bone, erosion of vestibule fundus, and absence of 1.3 and 1.4 dental units. Incisional biopsy confirmed AOT diagnosis. Treatment choice was surgical enucleation with curettage under general anesthesia. Dental units were removed due to impossibility of orthodontic traction. In 2nd postoperative year, regression of volume was observed. Radiographic exam showed bone neoformation with no signs of lesion recurrence. Due to the rarity of this tumor it is important that all cases are described to contribute to literature and statistical data.

Keywords: Tumor; Odontogenic Tumors; Biopsy; Treatment; Tooth

¹ Master degree student in dentistry. Department of Oral and Maxillofacial Surgery, Bahiana School of Medicine and Public Health, Salvador, Bahia.

² Pos-graduated student in Interactive Processes of Organs and Systems. Federal University of Bahia.

³ PhD in Human Pathology. Assistant Professor of Bahiana School of Medicine and Public Health, Department of Basic Science, Bahiana School of Medicine and Public Health, Salvador, Bahia.

⁴ PhD in Oral Pathology. Assistant Professor of Bahiana School of Medicine and Public Health, Department of Basic Science, Bahiana School of Medicine and Public Health, Salvador, Bahia.

⁵ PhD in Laser. Assistant Professor of Bahiana School of Medicine and Public Health, Department of Oral and Maxillofacial Surgery, Bahiana School of Medicine and Public Health, Salvador, Bahia.

Correspondence to:

Alena Ribeiro Alves Peixoto Medrado.
E-mail: alenamedrado@hotmail.com

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1 BACKGROUND

Adenomatoid odontogenic tumor (AOT) is a rare, benign, slow-growing tumor, comprising about 3% of all odontogenic tumors. Recently, it has been included among tumors derived from odontogenic epithelium with mature fibrous stroma without odontogenic ectomesenchyma¹⁻³. It is accompanied by increase in volume, which in most cases, is associated with lack of tooth eruption. Tumor more commonly affects maxilla than mandible (2:1), specifically, canines. There is greater predilection for female sex (2:1) between the 2nd and 3rd decade of life, and relapse is rare⁴⁻⁸.

AOT has three clinical-pathological variants: follicular variant that is associated with an unerupted tooth (73%); extrafollicular variant that usually presents between dental roots (24%); and peripheral variant (3%) that presents as small sessile masses in the vestibular gingival mucosa, which may also present slight erosion of the buccal cortical bone^{1,6,9,10}. Radiographically, AOT appears as a radiolucent lesion with well-defined contours, which is circumscribed by a well-defined, often delimited, cortical bone, and is usually associated with an unerupted tooth (follicular variant) or presents between dental roots (extrafollicular) with calcified points within lesion, with appearance of snowflakes^{6,7,9,11}. Histologically, this tumor is well-defined and has several arrangements of the odontogenic epithelium, i.e., it is grouped in structures similar to rosettes in the scarce connective tissue stroma. Amorphous eosinophilic material may present between cellular arrangements. Furthermore, pre-ameloblast-like epithelial cells can proliferate and cluster into niches, forming tubular structures called pseudoducts, which in some areas are filled with calcified material and in other areas are scarce or completely non-existent^{1,6,9,12,13}.

Treatment of choice is conservative surgical procedure with enucleation and curettage, because it is an encapsulated lesion with a non-aggressive behavior^{2,6}. Incisional biopsy and puncture aspiration are diagnostic procedures indispensable for surgical planning^{1,5,11-13}. Given the rarity of this lesion and the involvement of different structures in young patients, all affected cases need to be described. This study aims to report a clinical case of a pediatric patient diagnosed with follicular AOT in the right maxillary premolar region that was treated satisfactorily by surgical excision of the lesion.

2 CASE DESCRIPTION

An 11-year-old girl was accompanied by a care giver to a Stomatology Clinic in a Dentistry university

in 2015. Patient had increase in volume at right maxilla, with asymptomatic evolution of 1 year. As reported by the mother, patient did not have basic diseases and drug allergies and did not chronically use medications. Patient underwent an incisional biopsy that confirmed AOT.

Then, patient was referred to Oral and Maxillofacial Surgery Clinic of an University Hospital, where extra-oral physical examination showed a hardened volume increase, sensed by palpation, at the middle third of the right side of face, affecting canine pillar region. Intra-oral examination showed expansion of buccal cortical bone of the right maxilla, erosion of the right maxillary vestibule fundus, and absence of dental units 1.3 and 1.4. Patient denied experiencing pain and reported paresthesia in wing of the nose and right upper lip.

Panoramic radiograph showed radiolucent unilocular image with well-circumscribed radiopaque halo measuring about 3 cm in its largest diameter, with foci of intralésional calcification; associated with the crown of unit 1.4 causing impaction of unit 1.3 (Figure 1).

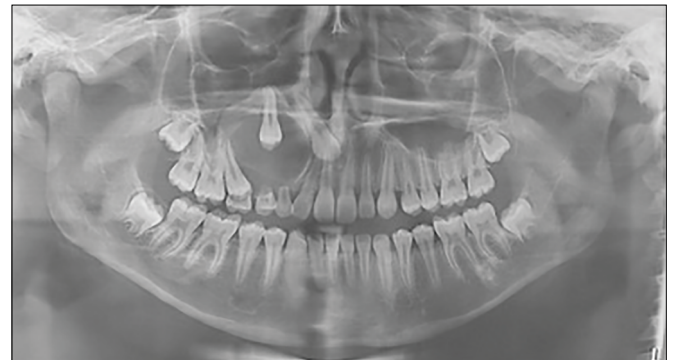


Figure 1. Panoramic radiograph showing unilocular well-circumscribed lesion involving unit 1.4. and the ectopic position of unit 1.3.

With the consent of the mother, treatment of choice was surgical enucleation with curettage under general anesthesia. Newman's access was performed with the opening of a bony window to lesion expose. Surgical specimen together with units 1.3 and 1.4 were removed due to impossibility of orthodontic traction (Figure 2). The microscopic examination of the lesion revealed fragments of odontogenic neoplasia that comprised fusiform epithelial cell masses that formed nests or rosettes and presented with the formation of small tubular structures and mineralized matrix area and a capsule of connective tissue in the periphery of the tumor with focal areas of hemorrhage and calcification (Figure 3).

Patient returned on the 7th postoperative day with a clean wound, no signs of dehiscence or infection, and no painful symptomatology. In the 2nd postoperative year, total

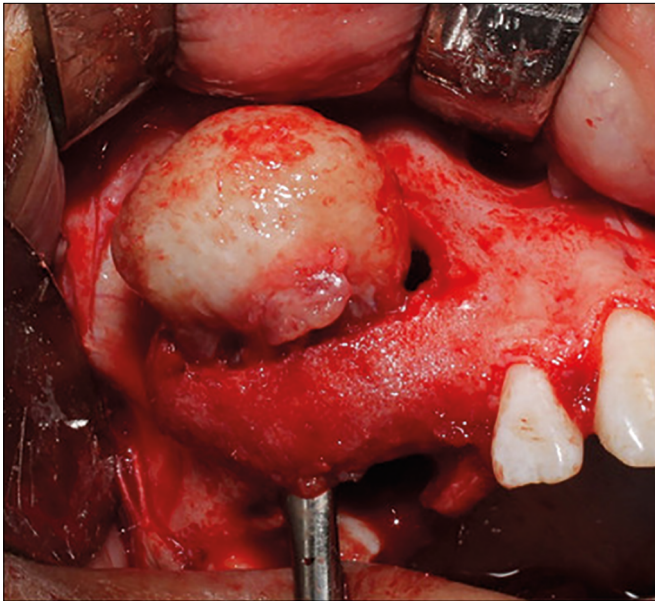


Figure 2. Bone window performed for enucleation of the lesion.

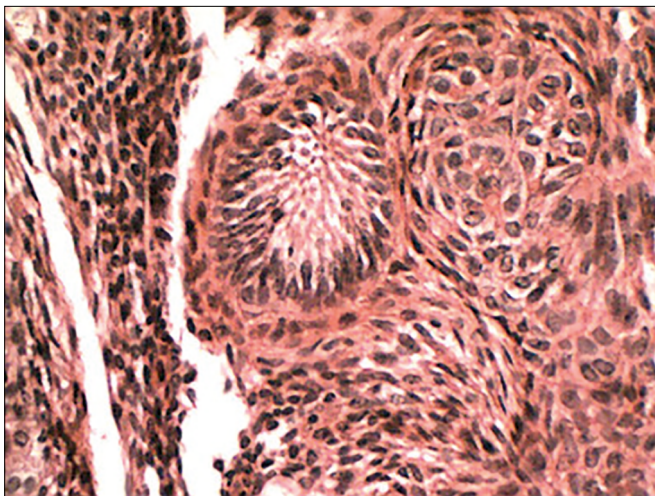


Figure 3. Presence of rosettes in the tumor parenchyma. Hematoxylin-Eosin, 400X.

eruption of unit 1.5 and regression of the volume increase due to the expansion of the vestibular cortex were observed. Radiographic examination showed neof ormation and bone remodeling with no signs of lesion recurrence (Figure 4).

3 DISCUSSION

Our study is in agreement with the literature, that says follicular type has a higher prevalence and women are more affected than men. Furthermore, this study increases the number of reported cases of AOT in maxilla, as current literature does not show a predilection for any of gnathic bones. Regarding involvement of



Figure 4. Panoramic radiograph in the 2nd postoperative year showing new bone formation.

impacted teeth, in present case, premolar unit 1.4 was involved in the lesion, which was not consistent with previously reported cases⁷.

An 11-year-old girl with a follicular AOT in the maxilla associated with the first unerupted premolar (1.4), caused the displacement and impaction of the canine (1.3). Despite the many characteristics that appear to be common among other AOT cases, the tooth involved in the lesion was a premolar. Unit 1.3 was also compromised because it was displaced by the lesion to such an extent that it was not possible to preserve it.

Although asymptomatic, patient had an increase in volume that affected her facial aesthetics and compromised her social life. Increase in lesion volume was due to the expansion of the buccal cortical bone of the maxilla^{3,12}. According to Erdur et al.⁵ in 2016, owing to the slow growing and asymptomatic characteristics of lesions, most pathological entities are accidentally discovered during routine radiographic examinations and generally have a size smaller than 3 cm in the greatest diameter. This still incipient approach is due to the increased access of the population to dental services and imaging tests. When lesions exhibit expansive but slow-growing behaviors, more time is available for detecting them. However, in spite of all these characteristics of accessibility and the slow evolution of this type of neoplasia, such growth altered the profile and facial symmetry of the present patient .

Radiographically, lesion described, provided a well-circumscribed unilocular image delimited by a scleral line without fenestration. AOT affected the entire dental unit 1.4, compromising all its bone support, generating a large displacement of unit 1.3, and rendering their eruption trajectory unfeasible. All these characteristics are similar to those of the follicular variant, as described in the literature^{3,6,11}.

Conservative surgical approach with enucleation and curettage is the first-choice treatment³⁻⁶. In cases of follicular AOT, the involvement of bone support of the tooth defines the feasibility of its maintenance. In present case, only curettage was sufficient to treat patient because after 2 years of clinical follow-up, new bone formation with no sign of relapse of the lesion was observed. Preservation of the teeth, however, was not possible, which caused mutilation of a pediatric patient, who will need oral rehabilitation in the future. In 2015, Bonardi et al.⁶ cautiously decided on taking a more conservative approach with the maintenance of a dental unit involved in lesion; this decision depended on some variants such as the extent and relationship of the lesion with the impacted tooth, degree of displacement of the involved tooth, and remaining bone support. In present case, lesion completely involved unit 1.4, with no sufficient bone remnant to enable its permanence. Furthermore, the lesion generated a large displacement of unit 1.3 toward the floor of nasal cavity, not allowing a viable rash trajectory after lesion removal, even with orthodontic treatment.

It is necessary to establish the differential diagnosis of AOTs from other lesions because depending on their biological behavior, type of treatment varies from conservative to aggressive. Follicular AOT should be differentiated from dentigerous cyst, ameloblastoma, and ameloblastic fibroma when no foci of intralésional calcification is observed and when there is calcification with calcifying epithelial odontogenic cyst, ameloblastic fibro-odontoma, calcifying epithelial odontogenic tumor, and fibro-bone marrow^{5,9}. Extrafollicular AOT should exclude the possibility of residual cyst and lateral periodontal cyst. Moreover, peripheral AOT should be differentiated from fibrous gingival or fibrous epulis^{11,12}.

4 FINAL CONSIDERATIONS

Surgical enucleation along with unerupted dental units removal, proved to be an effective therapy for the treatment of TOA in the present case. Patient had no lesion recurrence until the present moment and continues in preservation with the surgical team.

Compliance with Ethical Standards

Funding: No funding received

Conflict of Interest: All authors (1, 2, 3, 4 and 5) declares that he/she has no conflict of interest.

Ethical approval: Although this paper involves a person, as it is a case report, this article does not contain any studies with human participants or animals performed by any of the authors.

Informed consent: Informed consent was obtained from the patient's parent and it has been uploaded as we sent this manuscript.

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