

# Adenomatoid odontogenic tumor in the anterior mandible: a case report

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## Abstract:

Adenomatoid odontogenic tumor (AOT) is a rare, non-invasive, benign neoplasm of slow and progressive growth, originating from the odontogenic epithelium. The benign aspect of AOT predicts a more conservative approach through enucleation and curettage, with larger lesions being subjected to marsupialization prior to surgical treatment. This article aims to report a case of adenomatoid odontogenic tumor in the anterior mandible in a male patient who was treated with surgical marsupialization and posterior enucleation. Auxiliary therapeutic techniques can be used effectively to reduce the lesion, and accurate histological diagnosis is mandatory to avoid unnecessary aggressive surgery.

**Keywords:** Odontogenic tumors; Pathology; Differential diagnosis

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## INTRODUCTION

Adenomatoid odontogenic tumor (AOT) is a rare, non-invasive, benign neoplasm of slow and progressive growth, originating from the odontogenic epithelium<sup>1,2</sup>. At least, three variants of OAT have been described in the literature, follicular intraosseous, extrafollicular intraosseous and peripheral<sup>3,4</sup>. This tumor occurs predominantly in the second decade of life, in the anterior region of the maxilla and presents female predilections<sup>1,5</sup>. The follicular variant is known to be associated with an unerupted tooth, usually canine<sup>4,6</sup>. The lesion remains clinically imperceptible for a long time, however, it can produce displacement of adjacent teeth<sup>2</sup>, expansion of the surrounding bone and facial deformity in more aggressive cases<sup>4,7</sup>.

Radiographically, they are presented as well-defined unilocular radiolucent areas with varying degrees of calcification<sup>5,8</sup>. Histologically, the World Health Organization (WHO) defined AOT as a neoplasm of odontogenic epithelium with structures similar to ducts, with varying degrees of alterations in the connective tissue, as well as the presence of a well-developed connective tissue capsule<sup>3,6,9</sup>. The benign aspect of this tumor predicts a more conservative approach through enucleation and curettage<sup>10,11</sup>, and larger lesions may be previously submitted to marsupialization<sup>12</sup>.

The aim of the present paper is to report a case of follicular AOT in the anterior mandible of a male patient.

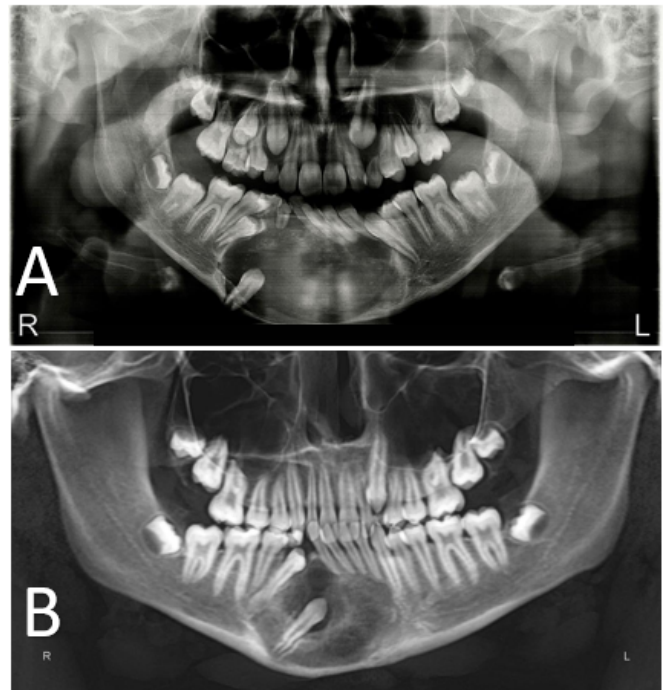
## CASE REPORT

A 12-year-old male patient attended the Surgery and Traumatology Service of the João de Barros Barreto University Hospital, complaining of a lump in his chin. On clinical examination, an increase in volume was observed in the anterior region of the right mandible, with firm consistency and painless on palpation, with an evolution of approximately 1 year. On radiographic examination, a well-defined unilocular radiolucent area was observed in the anterior region of the mandible, presence of diffuse calcifications in the interior of the lesion associated with tooth 43, in addition to root displacement of adjacent teeth (Figure 1A).

An incisional biopsy was performed for histopathological analysis, when spindle-shaped epithelial cells were seen forming rosette-like structures in a sparse fibrous stroma, thus confirming the diagnosis of follicular AOT. Marsupialization was performed for 20

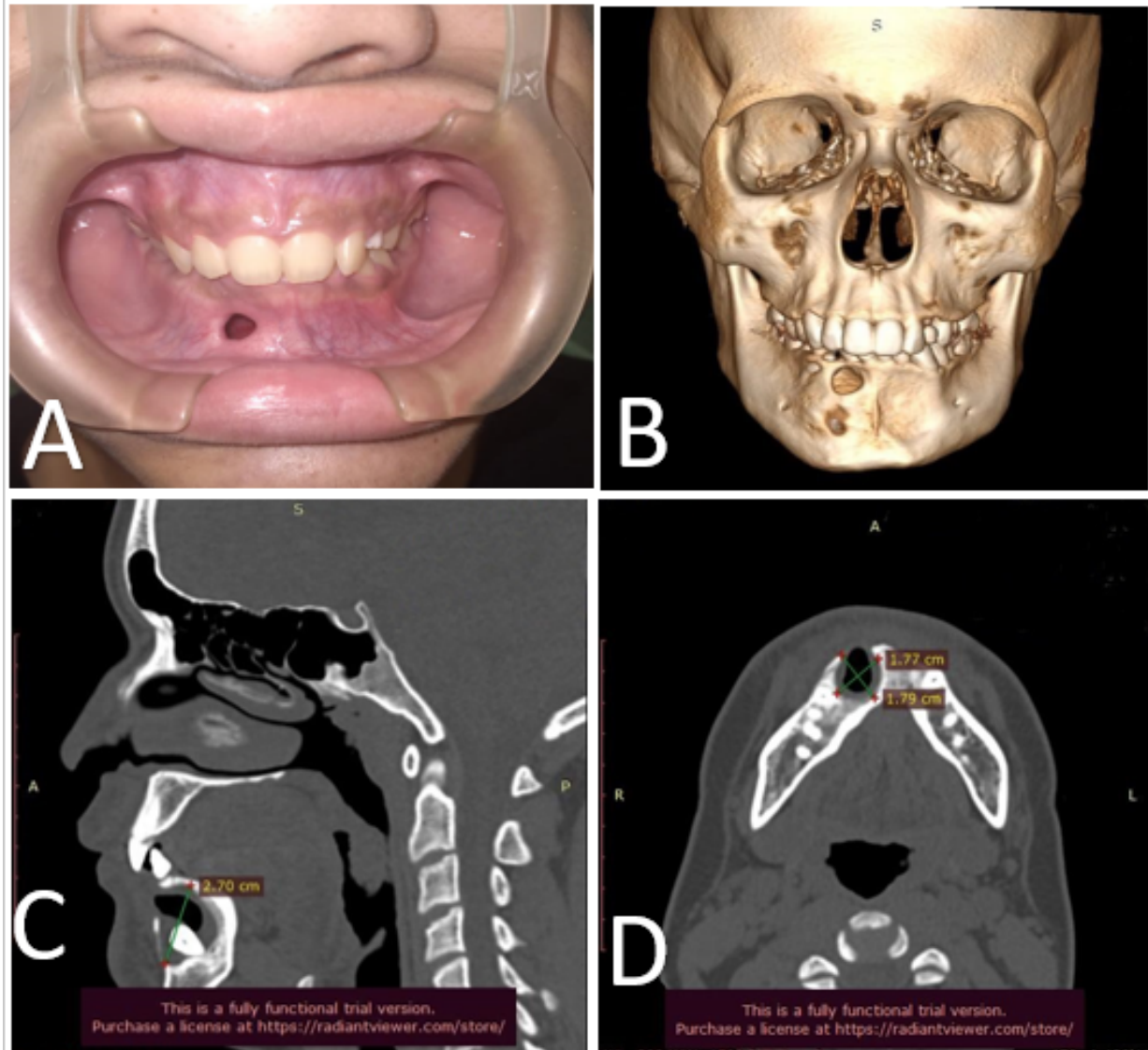
months and periodic clinical and radiographic follow-up with panoramic x-rays, which verified the progressive regression of the lesion (Figure 1B). After a significant reduction, a preoperative computed tomography of the face was requested for surgical planning, in which a well-defined lesion measuring 1.79 cm at its largest diameter was observed (Figure 2A-D).

The patient underwent a surgical procedure under general anesthesia for complete excision of the tumor, and the surgical specimen was sent for histopathological evaluation that confirmed the previous diagnosis (Figure 3A-C). Tooth 43, which was associated with the TOA, was removed in the same surgical act due to its excessive mobility.



**Figure 1.** Radiographic aspects. A) Initial radiographic appearance evidencing an unilocular radiolucent lesion in the anterior mandible with presence of diffuse calcifications, teeth 43 included and root displacement. B) Pre-surgical radiographic appearance showing regression of the lesion.

In the 7-day postoperative period, the patient evolved with dehiscence of the surgical wound and intra-oral purulent secretion drainage in the anterior region mandible. Irrigation and suture of the surgical access were performed. Amoxicillin 500mg of 8/8 hours for 7 days and 0.12% chlorhexidine mouthwash were prescribed. Orientation with general care was also realized. Actually, the patient is under regular follow-up, with no signs of local infection or recurrence of the lesion.



**Figure 2.** Clinical appearance and computed tomography (CT). A) Clinical appearance after the marsupialization procedure. B) Facial CT exposing the bone defect. C) Sagittal CT showing lesion with 2.70 cm in diameter. D) Axial CT showing a bone cavity measuring 1.77cm x 1.79cm.



**Figure 3.** Trans-surgical procedure. A) Exposure of the adenomatoid odontogenic tumor. B) Immediate appearance after enucleation. C) Enucleated adenomatoid odontogenic tumor.

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## DISCUSSION

AOT is described by several authors as an uncommon neoplasm that represents from 2% to 7% of all odontogenic tumors<sup>2,10</sup>. According to More et al.<sup>9</sup>, this lesion is usually small, and mainly affects the anterior region of the maxilla in young female patients<sup>7,11</sup>. It is consistent with the characteristics of the present case report, since the lesion were extended in the entire anterior region of the mandible. However, we presented a male patient, different of worldwide epidemiology.

Liu et al.<sup>14</sup> argues that this pathology is asymptomatic, of slow growth, painless and that rarely exceeds 3 cm in the higher diameter<sup>6,10</sup>. Genno et al.<sup>4</sup> adds that AOT discovery usually occurs in routine examinations when investigating the absence of a tooth<sup>5</sup>. Opposite to what has been reported in the literature, the patient in this case was referred with a swelling in the anterior mandible that, radiographically, exceeded the usual measurements of this tumor.

According to Mosavat et al.<sup>2</sup>, 75% of cases of AOT are the follicular variant associated with an unerupted tooth. According to Dwivedi et al.<sup>15</sup> and Katiyar et al.<sup>16</sup>, the follicular adenomatoid odontogenic tumor exhibits on radiographic examination a well-defined radiolucent unilocular area around or surrounding the roots of an unerupted tooth, with varied degrees of calcification in its interior<sup>3,17,18</sup>, corroborating with the findings of the presented case.

The literature establishes that AOT is a pathology that shows clinical and radiographic similarity with several others, including lateral periodontal cyst, odontogenic keratocyst, calcified odontogenic cyst, ameloblastoma, ameloblastic fibroma and, mainly, the dentigerous cyst<sup>2,14,17,19</sup>. As mentioned by Câmara et al.<sup>20</sup>, 77% of AOT cases are initially identified as a dentigerous cyst. In agreement with Sadasivan et al.<sup>10</sup>, the clinical and radiographic diagnosis become inconclusive, requiring the histopathological examination for diagnostic elucidation.

According to Lang et al.<sup>13</sup>, AOT is histologically presented with sparse fibrous stroma covered by a fibrous capsule, in which spindle-shaped epithelial cells are capable of forming wounds, or increases in spiral volumes and structures similar to rosettes<sup>1,4,20, 21,22</sup>. Kundoor et al.<sup>6</sup> mentions it also presents tubular or ductiform structures surround by a central space delimited by a layer of columnar or cubic epithelial cells with cells demonstrating polarized nucleus<sup>10,13,23</sup>. Small foci of calcification may also be observed<sup>24</sup>. In this

report, the histopathological examination is similar with the characteristics described above.

The treatment of AOT differs considerably from other odontogenic pathologies<sup>5,25</sup>. Due to its benign behavior, non-invasive and that does not infiltrate bone tissue<sup>4,18</sup>, the literature agrees that a more conservative approach is indicated through enucleation and curettage for the treatment of this tumor<sup>10,15</sup>.

Other authors also mention that the enucleation of the AOT, followed by removal of the involved tooth and curettage is an effective and widely recommended treatment<sup>4,11,25</sup>. Saini et al.<sup>17</sup> adds that the presence of the fibrous capsule favors this approach<sup>3,10</sup>. In the current report, this therapy was applied and proved to be able to surgically remove TOA without any evidence of complications, favoring an excellent prognosis, compatible with what is exposed in the literature<sup>4,15,17</sup>.

## CONCLUSION

Although the conservative approach is indicated in the literature for the treatment of AOT, an accurate histological diagnosis is mandatory to avoid unnecessary mutilating surgery. It is also concluded that the marsupialization technique can be used as an effective initial strategy that enables the reduction of the lesion, preservation of adjacent bone structure and less surgical morbidity.

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