

Case report

Surgical treatment of Adenomatoid Odontogenic Tumor: A 5-year follow-up



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ARTICLE INFO

Article history:

Received 11 April 2017

Accepted 26 June 2017

Available online 6 July 2017

Keywords:

Adenomatoid odontogenic tumor

Impacted tooth

Odontogenic tumors

Oral surgery

ABSTRACT

Adenomatoid odontogenic tumor (AOT) is a rare odontogenic tumor with limited growth and usually associated with an unerupted permanent tooth. It is a benign lesion, with a low rate of recurrence after surgical treatment. This is a case report of an 11-year-old female with an asymptomatic tumor growth on the maxilla and palatal displacement of the lateral incisor. A unilocular lesion surrounding the coronal impacted right upper canine and the displacement of premolars were observed radiographically. After incisional biopsy, the tumor was diagnosed as AOT. The enucleation of the lesion and the removal of the impacted canine was performed. Four months later, the patient had not shown any signs of recurrence and was referred for orthodontic treatment. New surgical procedure was performed to remove the gums that covered the premolars to promote their eruption. Five years later, the premolars are in position in the dental arch and there are no signs of recurrence of the lesion. (Rev Port Estomatol Med Dent Cir Maxilofac. 2017;58(2):126-131)

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Tratamento cirúrgico de Tumor Odontogénico Adenomatóide: Acompanhamento de 5 anos

RESUMO

O tumor odontogénico adenomatóide (TOA) é um tumor raro com crescimento limitado e geralmente associado a um dente permanente não erupcionado. É uma lesão benigna, com baixa taxa de recorrência após tratamento cirúrgico. Relato de caso de uma menina de 11 anos de idade com crescimento tumoral assintomático na maxila e deslocamento palatino do incisivo lateral. Radiograficamente, foi observada uma lesão unilocular em torno da coroa do canino superior direito impactado e deslocamento dos pré-molares. Após a biópsia inci-

Palavras-chave:

Tumor odontogénico adenomatóide

Dente impactado

Tumores odontogénicos

Cirurgia oral

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<http://doi.org/10.24873/j.rpemd.2017.07.018>

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sional, o tumor foi diagnosticado como TOA. A enucleação da lesão e do canino impactado foi realizada. Quatro meses depois, o paciente não apresentou sinais de recorrência e foi encaminhado para tratamento ortodôntico. Um novo procedimento cirúrgico foi realizado para remover a gengiva que cobria os pré-molares para promover sua erupção. Após cinco anos, os pré-molares encontram-se em posição no arco dentário e não existem sinais de recorrência da lesão. (Rev Port Estomatol Med Dent Cir Maxilofac. 2017;58(2):126-131)

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Introduction

Odontogenic tumors and hamartomas are included in a wide variety of rare lesions that originate from odontogenic tissues and are present at variables levels of differentiation. The determination of its exact nature (i.e., hamartoma or neoplasm) is difficult and often inconclusive, which makes the nomenclature for these types of lesions difficult.¹

The Adenomatoid odontogenic tumor (AOT) was first reported in 1907, called the pseudo-adenoameloblastoma.² Different nomenclatures have also been used to describe this tumor: adenoameloblastic and ameloblastic adenomatoid tumor. The term Adenomatoid odontogenic tumor, adopted by the World Health Organization classification of odontogenic tumors in 1971, is still the most accepted and currently used nomenclature.^{3,4}

AOT is a rare and non-aggressive epithelial odontogenic tumor that occurs in intraosseous and peripheral forms. The intraosseous variants are the most frequent and include follicular and extrafollicular types. Radiographically, the central lesions (intraosseous) appear as a unilocular and radiolucent area, which is well-defined and often associated with an impacted tooth. Two-thirds of intraosseous cases present radiopacity within them.⁵

This type of lesion corresponds to 2% to 7% of all odontogenic tumors, usually affecting young patients, mostly during their second and third decades of life. Women are affected more often than men (a ratio of 1.9:1.0), and the lesions tend to occur in the anterior maxilla region.^{4,6}

The aim of this study was to report the surgical management of an AOT, show the results from the five years of follow-up, and discuss clinical and radiographic aspects with the current literature.

Case report

An 11-year-old female patient was referred to the Oral and Maxillofacial Surgery service of the School of Dentistry of the Federal University of Minas Gerais (UFMG), Belo Horizonte, Brazil, presenting a 5-6-month history of asymptomatic tumor growth on the right side of the maxilla. There was no history of trauma and the lesion had progressively increased in size in recent months.

Clinical examination revealed swelling with ill-defined margins, with normal overlying mucosa, which was firm on

palpation. The buccal cortical plate of the right maxilla was expanded from the central incisor to the first molar and palatal displacement of lateral incisor on the same side (Figure 1). Facial asymmetry was not observed upon extra oral examination.

The panoramic radiography showed a well-defined, unilocular radiolucency in the maxilla associated with the unerupted right upper canine, with no evidence of calcifications or root resorptions. It also showed the displacement of first and second premolars on the same side and a delay in its eruption process (Figure 2). Radiographic and clinical findings were compatible with the diagnosis of dentigerous cyst, AOT, and unicystic ameloblastoma.

Fine needle aspiration was negative and an incisional biopsy was performed. Histological examination revealed 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, confirming the diagnosis of an AOT (Figure 3).

After confirming the diagnosis, the enucleation of the lesion was performed under local anesthesia, together with the removal of the impacted canine and deciduous molars (Figure 4). The final diagnosis of an intraosseous follicular variant of AOT was reconfirmed after the specimen had been microscopically examined.



Figure 1. Clinical image showing right anterior maxillary swelling with the expansion of the buccal cortical plate, palatal displacement of the lateral incisor, and the absence of upper right canines and premolars.

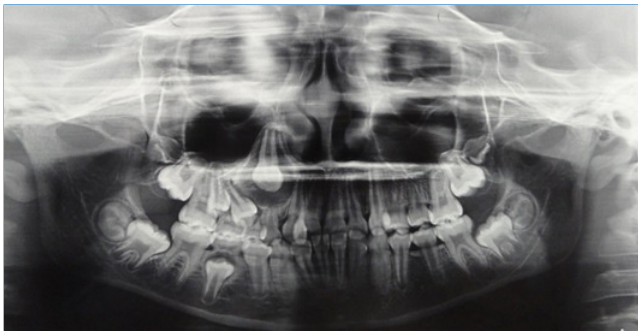


Figure 2. Panoramic radiography shows the unilocular lesion surrounding the coronal impacted right upper canine beyond the amelocemental junction and the displacement of the first and second premolars on the same side.

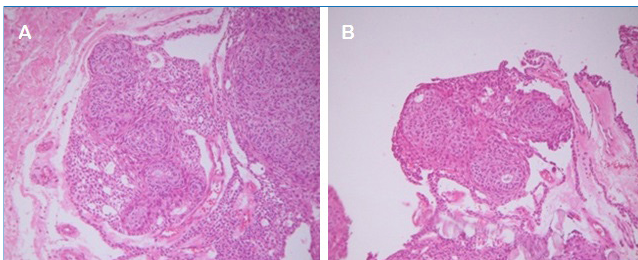


Figure 3. A: Higher power (x100) histology shows a hematoxylin and eosin stained section with a closer look at the spindle shaped epithelial cells arranged in whorls and strands. B: A duct-like structure lined by one layer of cuboidal epithelial cells.

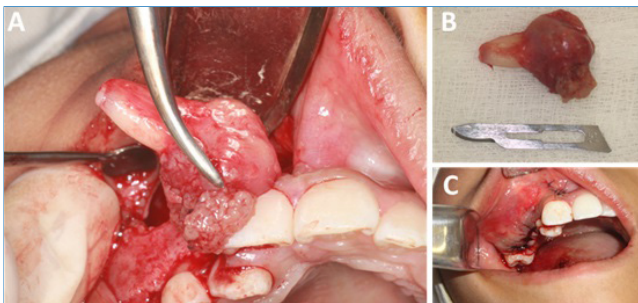


Figure 4. A: Enucleation of the lesion and associated impacted canine. B: Gross-examination of the specimen showed a single, well-encapsulated soft tissue. C: Immediate postoperative photograph.

Four months later, the patient showed no signs of recurrence and was referred for orthodontic treatment. A new surgical procedure was performed to remove the gums that covered the premolars and promote their eruption.

The patient underwent follow-up and, five years after surgery, has shown no signs of recurrence (Figures 5 and 6).

Discussion

AOT is a rare, hamartomatous, epithelial lesion of odontogenic origin.^{4,6} It's one of the most controversial lesions, due to its

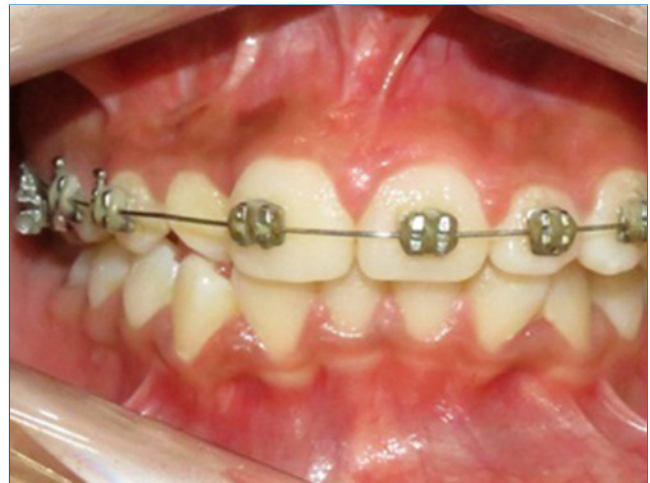


Figure 5. Five-year clinical postoperative follow-up showed no signs of recurrence.

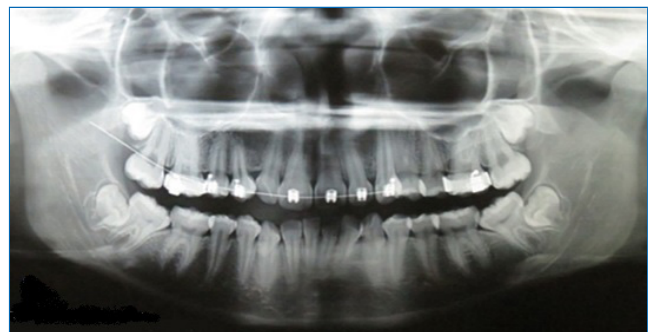


Figure 6. Five-year postoperative panoramic radiography shows normal healing and no signs of recurrence.

histopathological characteristics and similarity to ameloblastoma, thus previously receiving the name of pseudo-adenameloblastoma.² When compared to ameloblastomas, the most common odontogenic tumor, AOT is a nonaggressive tumor, encapsulated with limited growth and no tendency of recurrence. It is usually associated with an unerupted permanent tooth. Radiographically, in most cases, AOT shows a unilocular radiolucency with well-defined borders and may contain numerous dispersed radiopaque foci.^{4,7} Except for the absence of calcifications in the lesion, the present case presents the classic characteristics of the lesion.

Neoplastic or hamartous lesions can develop at any stage of a complex process called odontogenesis.⁸ In relation to the theory about the origin and pathogenesis of AOT, it seems that this tumor is derived from the odontogenic epithelium of the dental lamina complex or its cellular remnants situated in the gubernacular cord.⁹

The gubernacular cord is a fibrous innervated, vascularized, and lymphatic channel with epithelial cells or cell clusters from the fragmented dental lamina running in a bony channel called the gubernacular canal, which connects the pericoronal follicular tissue of the permanent tooth to the alveolar crest and the palatal gingiva of the deciduous tooth. Some authors conclude that dental lamina in the gubernacular cord of the

developing permanent anterior teeth, such as incisors, canines, or premolars, seems to be an embryonic source of more than 96% of AOT.^{9,10}

A critical review of all case reports of AOT described in the literature between the years of 2012 and 2017 was performed and 56 references were found. However, due to lack of data, 48 cases were included in the analysis.

This type of lesion occurs mainly in the second decade of life, and is rare in patients over 30 years of age.^{11,12} All lesions occurred in the second and third decades of life, except for two cases¹³⁻⁴⁸ one that affected a 10-year-old⁴⁹ and 50-year-old male patient.⁵⁰ Women are more commonly affected than men, at a ratio of 2:1.^{11,12} Of all the cases evaluated, 61.9% occurred in female patients,^{13-34,46-48} The present report shows a case of AOT in an 11-year-old female patient.

This lesion is commonly located in the anterior maxilla and rarely in the mandible. It usually surrounds the crown of unerupted teeth, and 60% of the cases are associated with an impacted canine⁵¹ as was illustrated in this report. Of all the cases evaluated, only 35.7% affected mandible.^{13,15-17,21,30,34,35,43-45,48,49,52,53} One of these cases presented the peripheral variant of the lesion de peripheral type,⁵³ and another was associated with a deciduous teeth.⁴⁹

The structure of the cyst, its insertion around the whole tooth, not only in the amelocemental junction of an unerupted element, and the size of the buccal cortical expansion were not so typical of a dentigerous cyst. However, there are case reports in dental literature of AOT arising in association with other lesions (Table 1); therefore, it requires an indispensable meticulous histopathological evaluation.^{54,55}

The management of this tumor should be surgical. Enucleation of the lesion and removal of the impacted tooth and simple curettage is recommended, although there is a report of the preservation of the associated tooth, together with surgical and orthodontic treatment.¹² Of the cases reviewed, one was treated with marsupialization with subsequent³⁹ enucle-

ation and in another there was preservation of the associated tooth 15. The prognosis is excellent and the recurrence rate is 0.2%.⁵⁵

Conclusion

The treatment of choice for ATO is its surgical removal by enucleation and removal of the associated tooth when present. Despite a good prognosis and a low risk of recurrence, a clinical and radiographic follow-up of patients should be performed. In addition, the patient should receive multidisciplinary treatment from various dental specialties in the rehabilitation of the space caused by the lost tooth.

Ethical disclosures

Protection of human and animal subjects. The authors declare that the procedures followed were in accordance with the regulations of the relevant clinical research ethics committee and with those of the Code of Ethics of the World Medical Association (Declaration of Helsinki).

Confidentiality of data. The authors declare that they have followed the protocols of their work center on the publication of patient data.

Right to privacy and informed consent. The authors have obtained the written informed consent of the patients or subjects mentioned in the article. The corresponding author is in possession of this document.

Conflicts of interest

The authors have no conflicts of interest to declare.

Table 1. Cases of Adenomatoid Odontogenic Tumor (AOT) associated with other lesion

Authors, Year	Site	Treatment	Other Lesion Associated
Sathyaranayana et al., 2017 ¹³	Anterior mandible	Excision	Unicystic Ameloblastoma
Naidu et al., 2016 (2 cases) ⁴⁶	Anterior maxilla	Not Related	Cemento-ossifying fibroma
Majumdar et al., 2015 ¹⁹ and Manjunatha et al., 2015 ²⁰	Anterior maxilla	Enucleation, tooth removed	Dentigerous cyst
Rezvani et al., 2015 ³⁷	Anterior maxilla	Enucleation	CEOT and focal cemento-osseous dysplasia
Acharya et al., 2014 ³⁹	Anterior maxilla	Marsupialization, enucleation and tooth removed	Dentigerous cyst
Shephard et al., 2014 ⁴²	Anterior maxilla	Enucleation, tooth removed and after histological examination peripheral ostectomy.	Keratocystic odontogenic
Yamazaki et al., 2014 ⁴⁸	Posterior of mandible	Enucleation	Ameloblastoma (true hybrid neoplasm)
Prakash et al., 2012 ³⁴	Anterior of mandible	Enucleation, tooth removed	Central ossifying fibroma
Singh et al., 2012 ⁴⁷	Anterior maxilla	Enucleation, tooth removed	Dentigerous cyst

CEOT: Calcifying epithelial odontogenic tumor

Acknowledgements

We offer our deepest thanks to the institutions that provided technical support for the development and implementation of this study.

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