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Adenomatoid odontogenic tumor: a case report with unusual clinical features

Tumor odontogênico adenomatóide: relato de um caso com características clínicas incomuns

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ABSTRACT

Adenomatoid odontogenic tumors (AOT) are benign, non-invasive and slow growing tumors. The tumor typically affects the young population, particularly in females, and the majority of cases is diagnosed in the second decade of life. There are three variants of AOT: follicular, extrafollicular, and peripheral. The present report illustrates a case of AOT with unusual findings, including a relatively large size arising within the mandible, causing the expansion of the buccal and lingual cortical bone. Orthopantomography revealed a well defined radiolucent lesion circumscribed by a radiopaque halo around the root of tooth 43, which caused the displacement of the roots of teeth 42 and 44. An incisional biopsy was performed and the surgical specimen was submitted for histophatological examination, which revealed the diagnosis of TOA. The treatment performed was a surgical excision of the lesion, and no complications or recurrence of the lesion have been noted after 1 year of follow-up.

Keywords: Mouth neoplasms; Odontogenic tumors; Mandible.

RESUMO

O tumor odontogênico adenomatóide (TOA) é um tumor benigno, não-invasivo e de crescimento lento. O tumor atinge tipicamente a população jovem, particularmente o sexo feminino, e a maioria dos casos é diagnosticada na segunda década de vida. Existem três variações do TOA: folicular, extrafolicular e periférico. O presente relato ilustra um caso de TOA com achados clínicos incomuns, incluindo o tamanho relativamente grande da lesão dentro da mandíbula, causando expansão das corticais ósseas vestibular e lingual. O exame radiográfico panorâmico revelou uma lesão radiolúcida bem definida e circunscrita por halo radiopaco próximo a raiz do elemento 43, o que causou o afastamento das raízes dos elementos 42 e 44. Uma biópsia incisional foi realizada e a peça cirúrgica removida foi submetida ao exame histopatológico, que revelou o diagnóstico de TOA. O tratamento consistiu na excisão cirúrgica da lesão, sem complicações ou recidiva da lesão após 1 ano de proservação.

Palavras-chave: neoplasias orais; tumores odontogênicos; mandíbula.

INTRODUCTION

Adenomatoid odontogenic tumors (AOTs) are benign, non-invasive lesions with slow, but progressive, growth that accounts for 2.2–7.1% of all odontogenic tumors[1]. AOT usually affects young patients, mostly during their second decade of life.[2,3] The tumors preferentially affects females (male:female ratio, 1:2)[1,4] and have the tendency to occur in the anterior maxillary region[1,3].

There are three variants of AOT: follicular, extrafollicular, and peripheral.[2] The follicular type is a central intra-bony lesion associated with an unerupted tooth, and accounts for about 70% of all cases.[1,2] The extrafollicular type is also an intra-osseous lesion, but unrelated to an unerupted tooth, and represents 25% of all AOTs.[1] The peripheral type is a rare form that arises in the gingival tissue, and only 18 well-documented cases have been reported.[2]

In the present paper we report a case of this uncommon odontogenic tumor of unusual size, and located in an unusual anatomical site, the mandible, with asymptomatic swelling and slow growth, and causing the divergence of roots.

CASE REPORT

A 13-year-old girl, a melanoderm, was referred to the dental service, presenting a facial asymmetry due to swelling of the mandible, which was otherwise asymptomatic. It was reported to be developing over a 2 year period (Figure 1). Intraoral examination revealed a tumor with a firm consistency, causing the expansion of the buccal and lingual bone corticals (Figure 2). The mucosal surface had an

erythematous aspect. Radiographic examination revealed a well-defined radiolucent lesion circumscribed by a radiopaque halo, surrounding the root of tooth 43 and rejecting the roots of teeth 42 and 44 (Figure 3). An excisional biopsy was performed, and the specimen was submitted for histopathological analysis. Microscopic examination revealed a lesion composed of sheets of variously-sized solid nodules of cuboidal and columnar epithelial cells, nests, and rosette-like structures containing eosinophilic droplets (Figures 4–6). Tubular duct-like spaces were also observed, lined by a single layer of columnar cells, with nuclei spread away from the lumen. There were also mineralized areas, and the stroma was scarce. With the clinical, radiologic, and histopathologic features, a final diagnosis of extrafollicular AOT was made.

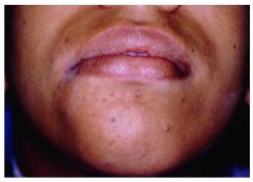
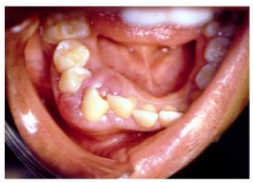


Figura 1



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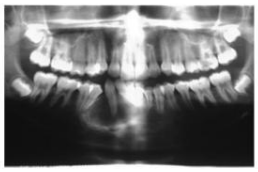

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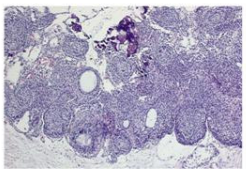
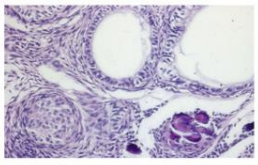


Figura 4



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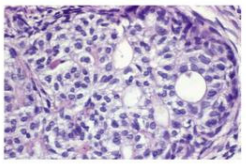

Figura 6

Figure 1 – Facial asymmetry due swelling of the mandible.
Figure 2 – Clinical aspect showing a firm, sessile lesion with an erythematous surface, and promoting the expansion of the buccal and lingual bone corticals.
Figure 3 – Orthopantomography showed a well-defined radiolucent lesion surrounding the root of tooth 43, circumscribed by and radiopaque halo.
Figure 4 – Microscopic features: duct-like spaces and nests of epithelial cells, and globules of acellular calcified material among the neoplastic epithelium (hematoxylin and eosin stain; original magnification x100).
Figure 5 - Odontogenic epithelial neoplasm exhibiting nests of cubic and spindle-shaped cells, duct-like structures lined by a single layer of low columnar cells, and mineralized material (hematoxylin and eosin stain; original magnification x64)
Figure 6 - Solid nodules of cuboidal epithelium arranged in sheets and rosettes, sometimes presenting an eosinophilic amorphous material inside. The stoma was scarce (hematoxylin and eosin stain, original magnification x200).

Complete surgical removal was the definitive treatment for this patient. After the final diagnosis, the parents of the patient were informed about the benign nature of the lesion. No complications or recurrence of the lesion have been noted after 1 year of follow-up.

DISCUSSION

AOTs are slow growing, benign lesions of odontogenic epithelial origin. They are regarded as benign neoplasms by most authors, although some have classified them as harmatomas.[5,6]

AOTs are odontogenic tumors whose origin is still controversial. Some, but not all, of the follicular types of AOTs may derive from the odontogenic epithelium of a dentigerous cyst. Dental lamina remnants probably represent progenitor cells for the peripheral type of this benign odontogenic tumor.[4,7] Following entrapment, these epithelial remnants proliferate in response to an unknown stimulus, giving rise to the lesion.[8] Furthermore, Malassez remnants found in the periodontal ligament may possibly give rise to an extrafollicular AOT.[7] It has been theorized by Philipsen et al. (1992)[9] that the complex system of the dental laminae, or its remnants, is the likely origin of the AOT mimicking a periapical radiolucent lesion of the maxillary incisor area.

The age distribution ranges from 3 to 82 years, with 90% being diagnosed before the age of 30 years.[4,6,10] It is more common in females than in males at a ratio of 2:1. The maxilla is affected more than the mandible, with the anterior part of the jaw being more frequently involved than the posterior part.[2,11] The tumor has been found to arise from the deciduous tooth-bearing area of the jaw. An impacted maxillary canine is the most common tooth to be associated with AOT.[12]

A recent worldwide literature survey has found the relative frequency of AOTs to be much higher than the 2–7% cited by Philipsen et al.[10] (1991). If the distribution of AOTs is analyzsed according to the geographic location, the relative frequency of AOTs in the different parts of the world is: Europe: 1–4%; Middle East: 2–4%; North America: 2–7%; South America: 4–7%; Asia: 1–16%; and Africa: 1–39%.[5].

The AOT occurs in intraosseus as well as in peripheral forms.[2] There are three

variants of AOT: (i) folicular, (ii) extrafollicular, and (iii) peripheral. The intrabony variants comprise the follicular and the extrafollicular type. The peripheral type arises in the gingival tissue, but is extremely rare. It may show a slight erosion of the alveolar bone cortex, but rarely produces radiographically detectable changes.[10]

The follicular type is a central intraosseous lesion associated with the crown of an embedded tooth – frequently a maxillary canine[1] – mimicking a dentigerous cyst.[13] This is considered the predominant form of AOTs (about 75% of all reported cases).[2,10] The radiolucency is well demarcated.[6,13] In approximately two thirds of the reported cases faintly detectable radiopaque foci have been found[14].

The extrafollicular type is an intraosseus lesion that is not associated with an unerupted tooth, and the well-defined, unilocular radiolucency is found between, above or superimposed upon the roots of erupted, permanent teeth.[14] The predominant radiologic picture of intrabony variants is that of a unilocular radiolucent lesion with well-demarcated borders. Others produced cloudiness in the maxillary antrum or a mixture of opacity and lucency.[10,15] Toida et al.[16] (1990) reported that the radiographs generally showed a well-demarcated radiolucency, with or without regular intralesional radiopacity. These features are in accordance with the findings of the present study.

An intraoral or extraoral swelling is the main symptom of AOT, and the swelling is usually painless and slow-growing[16]. Clinically, all variants of AOTs are characterized by slow, but progressive, growth, with few or no subjective symptoms. Cortical expansion is a common finding in central variants, while penetration of the cortical plate is unusual.[10] In one unusual reported case, the tumor consisted of two lobes, one of which was located in the maxillary bone, and the other was partially in bone and partially in soft tissue.[17] Displacement of neighboring teeth because of tumor expansion, as seen in the present case, is much more common than root resorption.[2]

AOTs generally do not exceed 1 to 3 cm in the largest diameter;[10] however, Lee, Lee and Hwang[8] (2000) reported a case that reached 4 cm in diameter, and that represented a classic follicular type. Geist and Mallon[18] (1995) also reported a large size lesion located in the anterior mandibular region. The lesion we have diagnosed was extrafollicular, and located in the anterior mandibular region, an uncommon presentation for an AOT, as reported by Philipsen et al.[5] (2007) and Gouvêa et al.[19] (2009).

The diagnosis of AOT for a round, unilocular, radiolucent, well-defined lesion located between the dental roots in the anterior mandibular region was one of our clinical choices. However, many other lesions are more frequent and present with similar radiographic features. These include inflammatory odontogenic cysts, noninflammatory odontogenic cysts, and other odontogenic tumors.[20,21,22]

The present case had the classic microscopic features of an AOT. Microscopically, AOT is composed of spindle-shaped, solid nodules, or cuboidal epithelial cells that are organized in nests or rosette-like structures. Also, strands with a trabecular or cribriform arrangement, and duct-like spaces are present.[4,5,22] The tumor has droplets of intercellular eosinophilic material composed of heterogeneous fibrils (thin collagen, electron-dense fibrils, and masses of amyloid filaments, with or without collagen fibrils), and varying amounts of calcified material.[4]

AOTs have a benign biologic behavior, are encapsulated, and virtually never recur.[2,5] Clinicians might consider the occurrence of extrafollicular AOT in the anterior mandible.[18] The treatment for the present case involved surgical removal and follow-up. The patient has been re-checked every 3 months for 1 year, and there has been no recurrence to date.

CONCLUSION

Although there are differences in the population regarding the types, frequency and distribution, adenomatoid odontogenic tumors are rare and usually do not recur. The importance of our case was in the unusual location of the lesion, reaching the jaw of the patient. The lesion was resected, and there were no signs of recurrence after one year of clinical follow-up. Based on the characteristics described, this case report can contribute to the diagnosis of other similar cases of AOTs.

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