

Journal section: Oral Surgery
Publication Types: Case report

doi:10.4317/jced.2.e149

A case report of desmoplastic ameloblastoma

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Received: 13/05/2010
Accepted: 17/06/2010

Márcio Bruno Amaral, Belini Freire-Maia, Marcella Rezende Serpa, Ricardo Alves Mesquita. A case report of desmoplastic ameloblastoma. J Clin Exp Dent. 2010;2(3):e149-52.
<http://www.medicinaoral.com/odo/volumenes/v2i3/jcedv2i3p149.pdf>

Article Number: 50308 <http://www.medicinaoral.com/odo/indice.htm>
© Medicina Oral S. L. C.I.F. B 96689336 - eISSN: 1989-5488
eMail: jced@jced.es

Abstract

Ameloblastomas are the most common benign neoplasm of odontogenic nature. Desmoplastic ameloblastoma (DA) was included in the World Health Organization Classification of Head and Neck Tumors (WHO-2005) as a variant of ameloblastoma with specific clinical, image, and histological features. This case report focuses on a DA which occurred in the mandible of a 25 year-old man. The main signals and symptoms included painless swelling with buccal expansion and tooth displacement. Panoramic and periapical radiographs and cone-beam computed tomography demonstrated an image mimicking a benign fibro-osseous lesion. The lower border of the mandible was not involved, but tooth displacement could be observed. An incisional biopsy was performed, and the DA was diagnosed. A marginal resection, maintaining the lower border of the mandible, was performed on teeth 46 to 31. The post-operative period was uneventful. The patient is undergoing routine follow-up and is currently free of disease.

Key words: Ameloblastoma, desmoplastic ameloblastoma, benign fibro-osseous lesions, odontogenic tumors.

Introduction

Ameloblastomas are the most common benign neoplasm of odontogenic nature that directly affects the jaws. Desmoplastic ameloblastoma (DA) was first reported by Eversole et al. (1) in 1984 and was recently included in the World Health Organization's Classification of Head and Neck Tumors (WHO-2005). DA is a tumor with specific clinical, image, and histological features. In addition, DA is a rare form of tumor and has been described in 115 cases in English literature (2).

Radiographically, 50% of DA shows an image suggesting a benign fibro-osseous lesion (2,3). Eventually, the clinical-radiographic features of DA may be ignored by surgeons and radiologists who more easily recognized the solid/multicystic variant of the ameloblastoma. Thus, to provide additional clinical-radiographic-histological data about DA, this article aims to describe a case of DA which occurred in the mandible of a 25 year-old man.

Case report

A 25-year-old man, with noncontributory cultural, social, and medical histories, was referred to the Department of Oral and Maxillofacial Surgery, Baleia Hospital/CEN-

TRARE in Belo Horizonte, Brazil, for evaluation and treatment of a tumor in the mandible region. The patient reported that the lesion had been present for five months. Through clinical examination, a swelling with asymmetry in the right parasymphiseal area could be observed extraorally (Fig. 1A). No cervical lymph nodes were palpable. However, a swelling of buccal gingiva and alveolar mucosa from tooth 42 to the tooth 45 could be observed intraorally. The lesion presented a firm consistency, coated by normal color and texture of the mucosa, measuring 35 x15 mm, which was painless and uneventful (Fig.1B). Teeth 41, 42, 43, 44, 45, and 47 were vital and immobile, but teeth 43 and 44 had been displaced. The panoramic and periapical radiographs revealed a poorly defined, mixed, radiolucent-radiopaque image, suggesting a benign fibro-osseous lesion. The displacement of the roots of teeth 43 and 44 (Figs. 2 A and B) could also be observed. Cone beam computed tomography (CBCT)



Fig. 1. A- Swelling produced an asymmetry in the right parasymphiseal area. B- Swelling of the buccal gingiva and alveolar mucosa from tooth 42 to tooth 45. The swelling is coated by a normal color and texture of the mucosa; teeth 43 and 44 are also displaced.

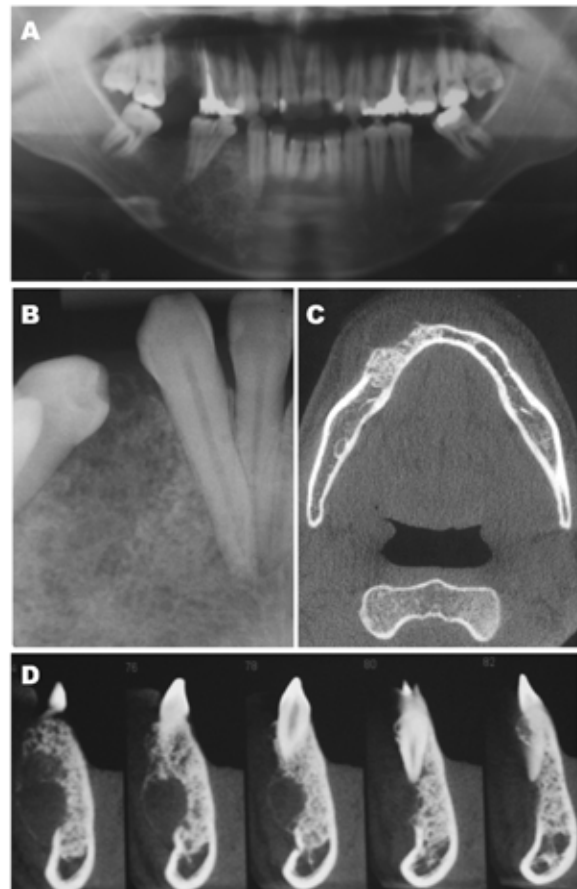


Fig. 2. A- Panoramic radiograph showing a poorly defined, mixed, radiolucent-radiopaque image, which is comparable to a benign fibro-osseous lesion. The roots of teeth 43 and 44 are displaced. B- Periapical radiograph image suggests a benign fibro-osseous lesion C- Cone beam computed tomography (axial plane) showing a mixed hyperdense-hypodense image with buccal expansion in the right anterior premolar region. D- In the parasagittal plane, a buccal expansion with no involvement of the lower border of the mandible can be observed.

scans were applied to better determine the extension and surgical planning for the lesion. The CBCT showed a mixed hyperdense-hypodense image with buccal expansion and the displacement of teeth 43 and 44 (Figs. 2C and D). Involvement of the lower border of the mandible (Fig. 2D) could not be observed. An incisional biopsy was performed and the DA was diagnosed (Figs. 3A and B). A segmental resection from teeth 46 to 31, maintaining the lower border of the mandible, was performed under general anesthesia. The treatment was performed through the mouth, and a reconstruction plate (2.4 mm - Neortho System, Curitiba, Brazil) was used to minimize the risk of fracture. The postoperative period was uneventful. The patient is currently undergoing routine follow-up with no signs of recurrence after 12 months.

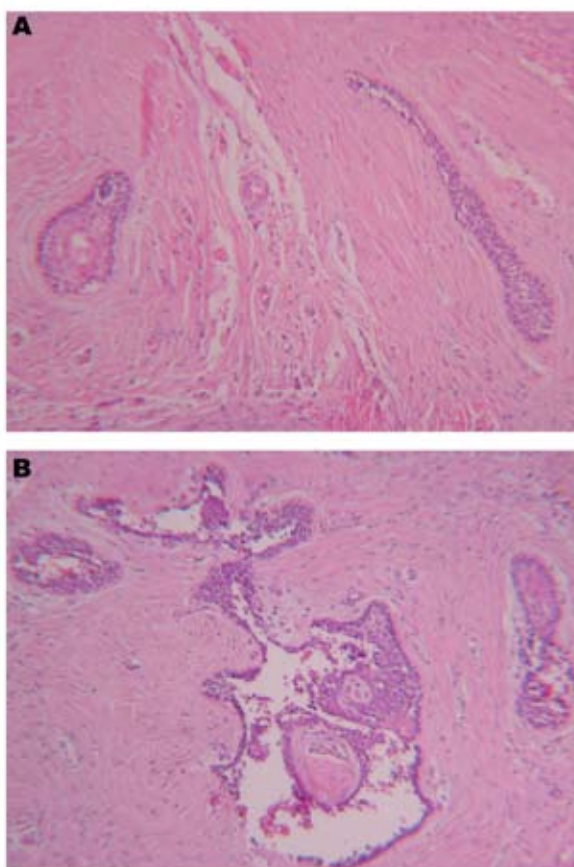


Fig. 3. A- Epithelial island and strand surrounded by a desmoplastic stroma (Hematoxylin and eosin, 200X original magnification). B- The epithelial islands appeared in irregular shapes (Hematoxylin and eosin, 200X original magnification).

Discussion

DA is a rare tumor, which, according to an English literature review carried out by Sun et al. (2) in 2009, can be found in 115 cases. It is a tumor with specific clinical, image, and histological features. The DA occurs more frequently in the 4th and 5th decades of life, and presents no predilection toward either gender. DA does

tend to affect the anterior premolar region of the jaws, but no prevalence between the maxilla and the mandible has been reported (2,3). As reported in prior literature, the present case of DA was also found in a young man, located in the anterior premolar region of the mandible, presenting painless swelling, buccal expansion and tooth displacement with no root resorption (4-6). The DA differs from the solid/multicystic ameloblastomas, which are more prevalent in mandibular molar/ramus regions, with variable sizes of swelling; pain tends to be rare, but root resorption of adjacent teeth is common (7,8).

In the image exams, solid/multicystic ameloblastomas appeared mainly as a multilocular radiolucency, similar to a soap-bubble or a honeycomb, with relatively well-defined borders (9,10). However, a well-defined radiolucent lesion may occur in a similar manner to that reported by Reichart et al. (11), where 51% of the tumors were described as unilocular. Nevertheless, the DA frequently appears as a poorly defined, mixed, radiolucent-radiopaque lesion mimicking a benign fibro-osseous lesion, especially when evaluating panoramic and periapical radiographs (2,7,12). Through the CBCT, however, it is possible to verify the tomographic limits of the DA. The present case report found images that are similar to other published cases of DA.

Histologically, solid/multicystic ameloblastomas contain two main histopathologic patterns: follicular and plexiform. These patterns consist of proliferating, irregularly shaped islands of the narrow cords of odontogenic epithelium embedded in a connective tissue stroma. By contrast, DA has appeared as irregularly shaped odontogenic epithelial islands surrounded by a narrow zone of loose-structured connective tissue embedded in desmoplastic stroma (9,13).

The management of solid/multicystic ameloblastomas and DA has been controversial due to the high rate of recurrence, especially in conservative treatments (enucleation and/or curettage) (14). A similar recurrence rate can be seen within the DA and solid/multicystic ameloblastomas (15). Sun et al. (2) (2009) identified a 15.9% rate of recurrence in DA cases treated by enucleation and/or curettage, with an average recurrence period of 36.9 months. The majority of DA cases reported in the literature have been treated by resection, most likely due to ill-defined borders, thus suggesting an infiltration process and an aggressive biological behavior, which has also been reported by some authors (3,15). In the present case report, the patient is currently showing no signs of recurrence.

The DA is considered a rare lesion, but information suggests that it can be considered as a distinct lesion. It is important to remember that new case reports regarding DA are important in an attempt to better define conclusions regarding its nature and biological behavior.

References

- 1.Eversole LR, Leider AS, Hansen LS. Ameloblastomas with pronounced desmoplasia. *J Oral Maxillofac Surg.* 1984;42:735-40.
- 2.Sun ZJ, Wu YR, Cheng N, Zwahlen RA, Zhao YF. Desmoplastic ameloblastoma – A review. *Oral Oncol.* 2009;45:752-9.
- 3.Ng KH, Siar CH. Desmoplastic variant of ameloblastoma in Malaysians. *Br J Oral Maxillofac Surg.* 1993;31:299-303.
- 4.Kawai T, Kishino M, Hiranuma H, Sasai T, Ishida T. A unique case of desmoplastic ameloblastoma: report of a case and a brief review of the English language literature. *Oral Surg Oral Med Oral Pathol Oral Radio Endod.* 1999;87:258-63.
- 5.Durmus E, Kalayci A, Ozturk A, Gunhan O. Desmoplastic ameloblastoma in the mandible. *J Craniofac Surg.* 2003;14:873-5.
- 6.Smullin SE, Faquin W, Susarla SM, Kaban LB. Peripheral desmoplastic ameloblastoma: report of a case and literature review. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2008;105:37-40
- 7.Manuel S, Simon D, RajendranR, Naik BR. Desmoplastic ameloblastoma: a case report. *J Oral Maxillofac Surg.* 2002;60:1186-8.
- 8.Wakoh M, Harada T, Inoue T. Follicular/desmoplastic hybrid ameloblastoma with radiographic features of concomitant fibro-osseous and solitary cystic lesions. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2002;94:774-80.
- 9.Philipsen HP, Reichart PA, Takata T. Desmoplastic ameloblastoma (including “hybrid” lesion of ameloblastoma). Biological profile based on 100 cases from the literature and own files. *Oral Oncol.* 2001;37:455-60.
- 10.Torres-Lagares D, Infante-Cossío P, Hernández-Guisado JM, Gutiérrez-Pérez JL. Mandibular ameloblastoma. A review of the literature and presentation of six cases. *Med Oral Patol Oral Cir Bucal* 2005;10:231-8.
- 11.Reichart PA, Philipsen HP, Sonner S. Ameloblastoma: biological profile of 3677 cases. *Eur J Cancer B Oral Oncol.*1995;31:86-99.
- 12.Kaffe I, Buchner A, Taicher S. Radiologic features of desmoplastic variant of ameloblastoma. *Oral Surg Oral Med Oral Pathol.* 1993;76:525–9.
- 13.Kishino M, Murakami S, Fukuda Y, Ishida T. Pathology of desmoplastic ameloblastoma. *J Oral Pathol Med.* 2001;30:35-40.
- 14.Nakamura N, Higuchi Y, Mitsuyasu T, Sandra F, Ohishi M. Comparison of long-term results between different approaches to ameloblastoma. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2002;93:13-20.
- 15.Keszler A, Paparella ML, Dominguez FV. Desmoplastic and non-desmoplastic ameloblastoma: a comparative clinicopathological analysis. *Oral Dis.* 1996;2:228–31.