CASE REPORT

Desmoplastic ameloblastoma: A case report

Tejavathi Nagaraj, Shamama Mumtaz, Soniya Kongbrailatpam, Ijum Doye

Department of Oral Medicine and Radiology, Sri Rajiv Gandhi College of Dental Sciences, Cholanagar, Bengaluru, Karnataka

Abstract

Desmoplastic ameloblastoma (DA) is an uncommon variant of ameloblastoma. In literature till now, much <150 sufferers have been suggested. DA accounts for about 4–13% of ameloblastoma, showing significant variations in anatomical site, imaging, and histologic appearance. We are presenting a case of DA in a 55-year-old female patient with a painless swelling in the anterior region of maxilla. Radiographic features of this lesion were indicative of ameloblastoma. An excisional biopsy confirmed it to a case of DA. The patient is on routine follow-up and is currently free of ailment.

Keywords:
Ameloblastoma, desmoplastic ameloblastoma, odontogenic tumor

Introduction

Ameloblastoma is the most common tumor after odontoma, it originates from the odontogenic epithelium.[1] It accounts about 1% of all cysts and jaw tumors and 18% of various odontogenic abscesses.[2] Among all the types of ameloblastoma, follicular and plexiform are the most common type, acanthomatous and granular are the second most common type of ameloblastoma. In recent years, the histomorphological spectrum of ameloblastoma has grown to include desmoplastic variants.[3] The first detailed report on the desmoplastic variation of ameloblastoma in English literature was given in 1984 by Eversole[4] who described the three cases and called them “ameloblastoma with the definite desmoplasia.” Classification of odontogenic categories by the World Health Organization (WHO) includes desmoplastic type as abnormal variation of ameloblastoma.[5] Ameloblastomas of 1–12% have been reported to be desmoplastic ameloblastomas (DA). All the other type of ameloblastoma mostly seen in the posterior region of the jaw but DA most often present in the anterior region of the jaw. The mixed radiopaque appearance represents the fibro-osseous lesion with well-defined boundaries.[6] All over the world, surgeons and radiologists are well aware of the clinical features of common ameloblastomas, however, they can ignore this variability.[7]

Case Report

A female patient of 55 years old came to the Oral Medicine and Radiology Department, Sri Rajiv Gandhi Dental College and Hospital, Bengaluru, with an asymptomatic swelling in her right maxilla that had started 1 month back. It was increasing gradually and reached to the present size. The swelling was asymptomatic. History of trauma was not positive her previous dental and medical record was not remarkable.

After examine the patient, physically asymmetry of the facial structure was noted because of the swelling on upper right side of face. Intraorally (Figure 1), a solitary bony hard non-tender and well-defined round to ovoid shape of swelling was present measuring approximately 4.0 cm in size, anteroposteriorly extending from the labial frenum to the distal of 13 and 2 cm superior-inferiorly extending from the attached gingiva of 12, 13 involving alveolar gingival and part of labial mucosa with overlying mucosa. Twelve, 13 showed drifting with marked extrudation, 12 associated with Grade 2 mobility. Palatal view (Figure 2) revealed a solitary well-defined round to ovoid shape of bony hard non-tender swelling of approximately 2 cm in diameter extending from midline to attached gingiva of 11, 12, 13.

Provisional diagnosis of ameloblastoma was given with differential diagnosis of calcifying odontogenic cyst, central giant cell granuloma. Periapical radiograph (Figure 3) and
Desmoplastic ameloblastoma: A case report

Nagaraj, et al.

Journal of Advanced Clinical & Research Insights ● Vol. 7:5 ● Sep-Oct 2020

Figure 1: Buccal extension of swelling

maxillary occlusal view [Figure 4] of region concerned showed large solitary diffuse multilocular radioluency with areas of calcification, with teeth displacement of 12. To rule out the superior extent or any other bony lesion, panoramic radiograph [Figure 5] was taken which showed no other bony involvement. Excisional biopsy was done which confirmed it DA. Histological section showed fibrous connective tissue with the desmoplasia with scanty inflammatory infiltrate. Lining epithelium showed ameloblastomatous feature with hyper chromatic nuclei. Few islands of follicles with few compressed follicles with peripheral ameloblast like cells and central stellate reticulum like cells are also seen with acanthomatous changes [Figure 6]. All details of the procedure were explained to the patient and a written informed consent was obtained. The involved teeth, that is, 12 and 13 were extracted. No recurrence has been observed in the 2 years of follow-up period.

Discussion

Ameloblastoma is a benign neoplasm derived from an enamel organ that usually shows aggressive behavior, causing severe expansion of the cortical bones and it may have high recurrence rate. Various histological subtypes of ameloblastoma have been described. The desmoplastic variant was given in detail in 1984 by Eversole et al. A swelling without pain and expansion of the bone is the most evident clinical feature in most of the...
cases consistent with this case report. The age at the start of the introduction is 42 years with the same age and same gender preferences as other ameloblastomas. According to the previous reports, DA lesions tend to be smaller (2 cm or less), with the exception of a single rare case that spreads to a larger culprit. This trend makes the DA different from normal ameloblastoma. In our case, the tumor appeared to originate near the right side of the maxillary lateral incisor and canine, because the roots of these teeth were distorted. In this case, there was root divergence but no periapical changes. Root resorption of adjacent teeth is commonly seen in normal ameloblastoma (92%) while it is rarely seen in DA (33%) indicating its indolent growth pattern.

DA shows more invasive characteristics compared to other types of ameloblastoma. This aggression may be because of: 1. The growth potential to increase in size. 2. A similar area on the upper jaw which leads to early spread on nearby surrounding areas. 3. Disseminated appearance of radiographic and histopathological findings of bone invasion.

DA has three radiographic changes that are described in the composition which are as follows: Type I (type of osteofibrosis) with a radiolucent and radiopaque appearance, Type II (type of radiolucency) which shows complete radiolucency, and Type III (mixed radiolucent and radiopaque) with mixed radiolucent and radiopaque appearance with more radiolucent changes. Radiological findings of this case reveal mixed radiopacities associated with type 1 osteofibrosis, which is considered as the most common type of radiographic changes compared to compound type which shows least common one. The DA lesion is characterized by osseous metaplasia within the dense septa and this may be the cause of the radiographic mixed opacity, which is not due to the mineral production in this tumor. The DA lesion is distinguished from the bone metaplasia inside the dense fibrous septa, so this can be the cause of mixed radiological appearance, maybe it is not due to calcified products of the tumor.

The radiology and the histology features of defective encapsulation and irregular borders confirm a radical surgical approach along with long-term follow-up.

Histopathology is the gold standard for diagnosing such lesions. If the biopsy sample find insufficiency to confirm the existence of tumour of ameloblastic layer can result to mislead diagnosis of DA as different odontogenic tumor. Not all ameloblastomas behave aggressively so it is important to differentiate between clinical types of ameloblastoma to provide appropriate treatment to patients.

**Conclusion**

DA is characterized by specific clinical features, imaging, and histological features. A good understanding of such cases is essential for deep analysis and long-term follow-up. The clinician should be careful about the rare presentation of the disease and should include DA as a differential diagnosis for any lesion ranging from a simple tumor to any fibro-osseous/neoplastic growth that introduces the anterior maxilla/mandible. A complete diagnosis requires histopathological examination. And with the power of recurrence, such cases should be treated with the complete resection.

This case is different from the other cases:
1. Usually, the involved teeth will show root resorption with extrudation, but in our case, there was extrudation without root resorption of the involved tooth.
2. In our case, there was no involvement of nasal floor unlike to other DA cases.

**References**

1. Lamichhane NS, Liu Q, Sun H, Zhang W. A case report on desmoplastic ameloblastoma of anterior mandible. BMC Res Notes 2016;9:171.
2. Sheikh S, Pallagatti S, Singla I, Kalucha A. Desmoplastic ameloblastoma: A case report. J Dent Res Dent Clin Dent Prospect 2011;5:27-32.
3. Thompson IO, van Rensburg LJ, Phillips VM. Desmoplastic ameloblastoma: Correlative histopathology, radiology and CT-MR imaging. J Oral Pathol Med 1996;25:405-10.
4. Eversole LR, Leider AS, Hansen LS. Ameloblastomas with pronounced desmoplasia. J Oral Maxillofac Surg 1984;42:735-40.
5. Kishino M, Murakami S, Fukuda Y, Ishida T. Pathology of the desmoplastic ameloblastoma. J Oral Pathol Med 2001;30:35-40.
6. Nair PP, Bhat GR, Neelakantan S, Chatterjee R. Desmoplastic ameloblastoma of mandible. BMJ Case Rep 2013;2013.
7. Sun ZJ, Wu YR, Cheng N, Zwahlen RA, Zhao YF. Desmoplastic ameloblastoma-a review. Oral Oncol 2009;45:752-9.
8. Katti SS, Katti SP. Desmoplastic ameloblastoma-a case report. Ann Clin Case Rep 2019;4:1695.
Craniomaxillofac Surg 1991;19:323-7.
10. Gardner DG. Some current concepts on the pathology of ameloblastomas. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1996;82:660-9.

How to cite this article: Nagaraj T, Mumtaz S, Kongbrailatpam S, Doye I. Desmoplastic ameloblastoma: A case report. J Adv Clin Res Insights 2020;7(5):82-85.