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Desmoplastic Ameloblastoma – A Case Report

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Abstract:

Desmoplastic Ameloblastoma (DA) is a rare variant of ameloblastoma. Cases of DA deserves special attention because of its rarity and high chance of misdiagnosis. We report a case of DA in a 45-year old female that presented as a painless swelling in lower anterior region. This report is an attempt to help the dental community in developing familiarity with the clinical and histological presentation of DA and at the same time advocating to develop a suspicion in recognizing such cases at right time.

Keywords: *Desmoplastic Ameloblastoma, Odontogenic tumor, Mandible, Anterior*

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1. Introduction

The Ameloblastoma is a true neoplasm of enamel organ type of tissues which does not undergo differentiation to the point of enamel formation. Robinson aptly described ameloblastoma as being a tumor that is 'usually unicentric, nonfunctional, inconsistent in growth, anatomically benign and clinically persistent'. The term Ameloblastoma was coined by Churchill in 1934. The ameloblastoma includes several clinical, radiographic and histological different types. Desmoplastic Ameloblastoma is rare, accounting for approximately 4% to 13% of ameloblastomas. [1]

It was first described by Eversole et al in 1984 as a new type of ameloblastoma which affected different mandibular areas, presenting a unique histopathological pattern and clinico-radiographic findings. They called it an 'Ameloblastoma with pronounced Desmoplasia'. [2]

This variation has been included in the World Health Organization's histopathological classification of odontogenic tumors as a rare variation of ameloblastoma.[3]

2. Case Report

A 45-year-old female patient reported to the outpatient department for complaint of painless swelling in lower anterior region for 2 years. Past surgical history revealed

previous surgery in the same region before two years for Dentigerous cyst that was treated by marsupialization.

Intra oral examination revealed a swelling extending from 34 to 45 region. On palpation, the swelling was non-tender and hard. The swelling was bony hard in consistency, non-fluctuant, non-reducible, non-compressible and non-pulsatile. Displacement of mandibular incisors were noted.

Orthopantomogram showed an impacted tooth surrounded with an ill-defined mixed radiolucent-radiopaque lesion extending from 34 to 45. There was evidence of root resorption, tooth displacement and ill-defined radiopaque borders. (Figure 1)

Clinical diagnosis of Odontogenic lesion was made and incisional biopsy was done for histopathological confirmation.

H & E stained section showed irregularly shaped odontogenic islands surrounded by desmoplastic stroma. The overlying epithelium was parakeratinized stratified squamous in nature. Foci of mild inflammation were also seen. (Figure 2)

Correlating the clinical and histopathological findings, a diagnosis of Desmoplastic Ameloblastoma was given. The patient was advised surgical resection and regular follow-up.

3. Discussion

DA is a rare variant of ameloblastoma, with an incidence of 4% to 13% among reported ameloblastoma cases [1]. Mintz et al reported that Desmoplastic ameloblastoma exhibits a more aggressive behavior than other types of ameloblastoma. This aggressiveness may be due to 1) potential to grow to a large size; 2) the common location in the maxilla leading to an early invasion of adjacent structures; 3) the diffuse radiographic appearance, and 4) histologic finding of bone invasion.[4]

A painless swelling or bony expansion are the most frequent clinical manifestation in most cases. The mean age of DA patients at the initial presentation ranges from 40 to 49, and DA exhibits a similar gender distribution to other ameloblastomas. Approximately 50% of DA occur in the maxilla, with the vast majority arising in the anterior or premolar portion of the jaws, which is not consistent with the usual location of conventional ameloblastomas [4-7].

It is suggested that desmoplastic ameloblastoma develops from the periodontal membrane of the related tooth. Moreover, some suggest that desmoplastic ameloblastoma might arise from epithelial rests of Malassez in the periodontal membrane. In this case, disappearance of the lamina dura and the periodontal ligament space of the involved tooth root was clearly identified.[5]

Tooth displacement is a common feature in desmoplastic ameloblastoma in almost 92% of the cases and root resorption is seen in just 33% of the cases. [8] The patient described in this report presented without root resorption, but displacement of the adjacent teeth and also the tumor was associated with a missing tooth.

In the literature three radiological presentations of DA are mentioned which are as follows: type I (osteofibrosis type) which has radiolucent as well as radiopaque appearance; type II(radiolucent type) which has a completely radiolucent appearance; and type III (compound type) which has radiolucent as well as radiopaque appearance combined with a large radiolucent change [9,10].

The management of DA has created a huge controversy due to its high recurrence

rate of approximately 36.9 months, especially after conservative surgery including enucleation or curettage. [11] Our patient was advised surgical resection to manage this entity.

4. Conclusion

We present a rare case of Desmoplastic Ameloblastoma in this case report. We need to summarize all the reported cases of DA along with its clinical and post-operative findings to ascertain its behavior.

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Figures & Legends:

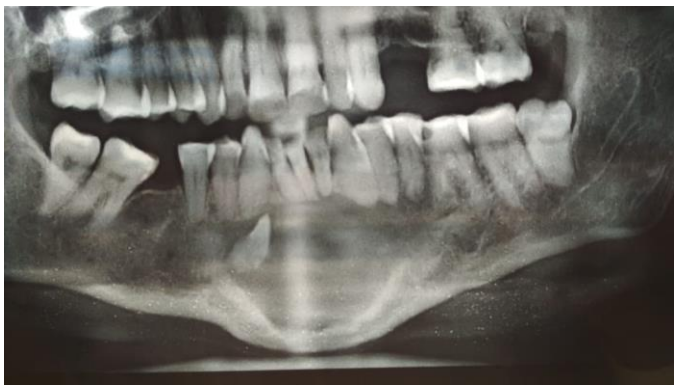

Figure 1: the orthopantomograph showing mixed radiolucency seen from 34 to 45 region with radio-opaque borders

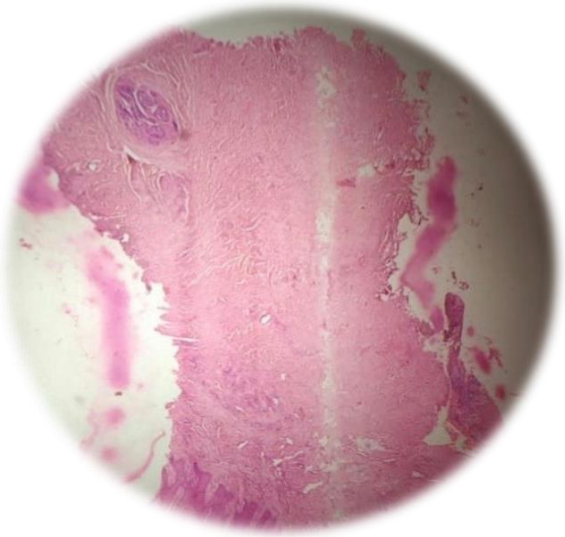

Figure 2: the photomicrograph depicting dense, fibrous connective tissue with compressed odontogenic islands (H & E 20x)