Case Report

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# Desmoplastic Ameloblastoma: A Case Report.

**Abstract:**
Desmoplastic ameloblastoma is defined as locally invasive, intraosseous and a rare variant of ameloblastoma which show slight male predilectionand its prevalence was within the fourth and fifth decades of life. Desmoplastic ameloblastoma (DA) is characterized by specific clinical, imaging, and histological features. We reporteda case of DA in a 28-year-old male with a painless swelling in the right anterior maxillary region. DA has greater predilection for maxilla with marked tendency for anterior regionas compare to other variant of ameloblastoma which can causeexpansion of lesion into maxillary sinus, high recurrence rate.Proximity to important anatomical structures like pterygopalatine fossa and orbitdemands extra caution in the approach during treatment planning.

**Keywords:** Desmoplastic ameloblastoma, anterior maxil

## Introduction:

Ameloblastoma is commonest benign odontogenic tumour of the jaws that constitute about 1% of all cysts and tumors of the jaws.[1] Usually ameloblastoma is slow growing and locally aggressive tumour causing expansion of the cortical bone, perforation of the lingual or the buccal cortical plate and infiltration of the soft tissues. Peak incidence of ameloblastoma is third and fifth decade of life but can be found in any age group with equal gender predilection (1:1) [1]. Ameloblastoma is sub classified into six histopathological subtypes; desmoplastic ameloblastoma is a rare histopathological subtype of ameloblastoma. Tumour shows a predilection for the anterior maxilla [2] and occurs most commonly within the fourth and fifth decades of life[3]. It accounts for 4%-13% of all ameloblastoma [4] and typically presents as painless, expansile mass. The radiographic features of DA differ markedly from other ameloblastoma variants. Radiographically, DA either presents as a mixed radiolucent/radiopaque lesion like benign fibro-osseous lesion or presents with multilocular radiolucencies that have ill-defined or well-defined borders [5]. It mostly shows cortical expansion and tooth displacement, but it rarely shows root resorption. Histologically, characteristic features include the presence of abundant, densely collagenous stroma containing small nests and strands of odontogenic epithelium[1].

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## Case report:

A 28 year old male visited the Outpatient Department of Oral Medicine & Radiology with the chief complaint of painless swelling of gums in upper front teeth region since three month. Patient was apparently all right 3 month back then he noticed slight bulge in upper front teeth region which gradually increased to present size. His past medical, dental and family history were non-contributory. On general physical examination, patient was systemically well with no constitutional sign and symptoms. His vitals were within normal range. On extraoral examinationthere was no significant facial asymmetry noticed, and no significant lymphadenopathy reported. Intraorally there was ill defined swelling of approximately 2x2.5 cm in size extending mesiodistally form right maxillary central incisor to right maxillary first premolar present labially(fig1), and also

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extending palatally in relation to right maxillary central incisor, lateral incisor and canine(fig 2). Over laying mucosa appear normal and,swelling was hard and non tender on palpation.Pulp vitality test showed positive response to the teeth in involved area. Based on the clinical finding, a provisional diagnosis of central giant cell granuloma was made. In clinical differential diagnosis lateral periodontal cyst, calcifying epithelial odontogenic tumour, ameloblastoma, squamous odontogenic tumour were included.

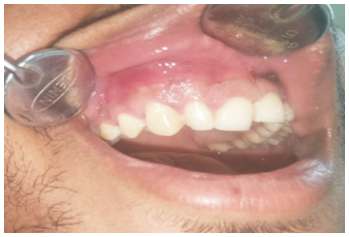

Figure 1: Swelling extending from 11 to 14.

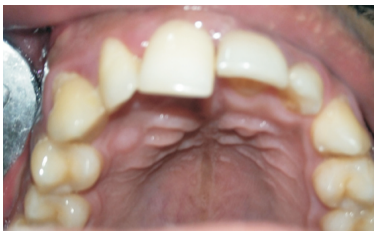

Figure 2: Intraoral Diffuse swelling involving right anterior hard palate in relation to 11, 12 and 13.

The routine blood investigation and serum level of calcium, phosphorus, alkaline phosphatase, PTH were within normal limit.A periapical radiograph of right maxillary lateral incisor and right maxillary canine region was taken which showed ill defined mixed radiolucent and radiopaque lesion with loss of laminadura and displacement of adjacent teeth. Anocclusal radiograph showing ill-defined mixed radiolucent and radiopaque lesion involving right anterior maxilla extending up to right maxillary first premolar with expansion of labial cortical plate and displacement of root of right maxillary central incisor, lateral incisor and canine labially, faint internal septation give partial locular appearance to the lesion (fig 3).Panoramic radiograph showing radiolucent lesion with displacement of root of right maxillary lateral incisor mesially and right maxillary canine distally (fig 4). Based on the radiographic finding differential diagnosis of fibro-osseous lesion like fibrous dysplasia, ossifying fibroma, periapical osseous dysplasia and calcifying odontogenic cyst, ameloblastoma was considered. Since lesion was small surgical excision performed under local anaesthesia and specimen was sent for histopathological examination to establish a definitive diagnosis.Histologically, the feature consist of dens collagenous stroma scattered in which strands, small Island and aggregate of neoplastic odontogenic epithelial cell with peripheral polisading of ameloblastomatous cells are noted. Over all histological features were suggestive of desmoplastic ameloblastoma(figure 5).

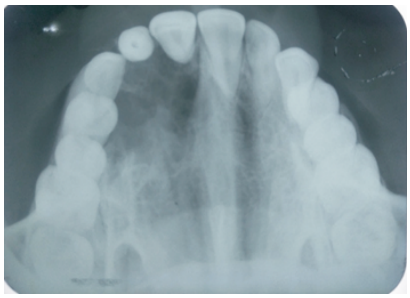

Figure 3: Occlusal radiograph showing ill-defined mixed radiolucent and radiopaque lesion involving right anterior maxilla extending upto 14 with expansion of labial cortical plate and displacement of root of 11, 12 and 13 labially. Faint internal septation give partial locular appearance to the lesion.

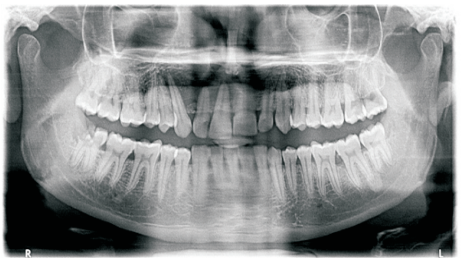

Figure 4: Panoramic radiograph showing radiolucent lesion with displacement of root of 12 mesially and 13 distally.

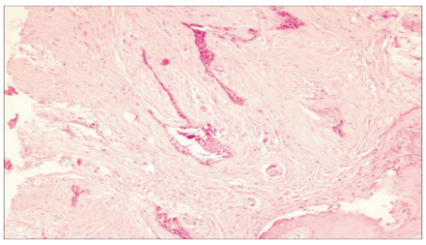

Figure 5: Histopathological picture shows dens collagenous stroma scattered in which strands, small Island and aggregate of neoplastic odontogenic epithelial cell with peripheral polisading of ameloblastomatous cells are noted.

## Discussion:

Ameloblastoma wasdescribed by Robinson in 1937 as a benign tumor that's “usually unicentric, nonfunctional, intermittent in growth, anatomically benign and clinically persistent.” It is a neoplasm of odontogenic epithelium[6],

principally of enamel organ-type tissue that has not undergone differentiation to the purpose of hard tissue formation. It is a slow-growing but locally invasive tumour. Peak incidence of ameloblastoma is in the third to fifth decades of life [7]. WHO and the International Agency for Research on Cancer, 2003, classified ameloblastoma as a benign tumour with odontogenic epithelium, mature fibrous stroma and without odontogenic ectomesenchyme. Ameloblastoma is again classified into: Solid/multicystic, Extraosseous/peripheral, Desmoplastic ameloblastoma, Unicystic. Six histopathological subtypes of solid ameloblastoma include follicular, plexiform, acanthomatous, basal cell, granular and desmoplastic. Mixtures of various histological patterns are commonly observed, and therefore the lesions are frequently classified supported the predominant pattern present.

In 1984 Desmoplastic ameloblastoma was first reported by Eversole et al. and it was recently included within the WHO's classification of head and neck tumours (WHO-2005).This tumour presented by an unusual histomorphology, including extensive stromal collagenization or desmoplasia, resulting in the proposed term ameloblastoma with pronounced desmoplasia or DA [1]. Desmoplastic ameloblastoma present a separate clinical and pathological feature because it differs from the other types of ameloblastoma in its anatomical location, morphology, and radiographic appearance. However, age and sex distribution don't differ from the other types of ameloblastoma. This tumour has a predilection for occurrence in the anterior or premolar region of the maxilla or the mandible. Ameloblastoma of maxilla tends to have a higher local recurrence rate compared with ameloblastoma in the mandible, because the thinness of the cortical bone in the maxilla is a less effective barrier to tumour invasion. The proximity of important anatomical structures demands extra caution within the approach during treatment planning. These include the pterygopalatine fossa and the orbit. These issues may contribute to the increase recurrence rates shown for the maxillary lesions (14.8%) in comparison to the mandibular lesions. It produces mixed radiolucent -radiopaque lesion with diffuse border that indicates that the tumour is more aggressive than other variants of ameloblastoma[8]. DA histologically appears as irregular odontogenic epithelial islands surrounded by a narrow zone of loose connective tissue embedded in desmoplastic stroma[9]. Recurrence rate of about 15.9% has been reported in DA cases treated by enucleation and/or curettage, with an average recurrence period of 36.9 months[4]. Malignant Transformation of a Desmoplastic Ameloblastoma to Squamous Cell Carcinoma is also reported in literature [10]

## Conclusion:

Clinician must be aware regarding the rare presentation of this benign tumor and DA must be included in differential diagnosis of mass/ growth occurring in anterior region of jaw. Radiological and histological finding of poorly incapsulation and ill defined border suggestive of infiltrative nature recommend complete resection as a treatment of choice and long term follow up.

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