



## Desmoplastic Ameloblastoma

### KEYWORDS

Benign Odontogenic tumor, Desmoplastic ameloblastoma, Odontogenic cyst.

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**ABSTRACT** *Desmoplastic ameloblastoma (DA) is benign, locally invasive, intraosseous infiltrative tumor with a relatively rare histological variation. DA has a nearly equal male to female ratio with prevalence within the fourth and fifth decades. This case report focuses on a DA which occurred in a 17 years girl with painless swelling with the involvement of midline to angle of mandible which deserves special importance because of its unfamiliar appearances, potentially aggressive nature and high chances of misdiagnosis. Computed tomography demonstrated ill-defined radiolucent lesion mimicking a dentigerous cyst. After performing incisional biopsy and histopathology showed dense, collagenous fibrous tissue in which the neoplastic epithelium forms small irregular islands having slender, straggling extensions. The final histological diagnosis was DA. Resection of mandible was a line of treatment. This article aims to provide additional clinical, radiographic and histological data about DA and to assist the dental community in better understanding of this variation in odontogenic tumor.*

### Introduction

Desmoplastic ameloblastoma is a tumor arising from odontogenic epithelium, despite their locally destructive nature, it is considered benign. The desmoplastic variant differs from other histologic types of ameloblastoma in that it is located in the anterior or premolar regions of the maxilla or mandible and its radiographic appearance is often more typical of a fibro-osseous lesion. Because only a few cases have been reported, the true biologic profile is yet to be fully understood. Desmoplastic ameloblastoma (DA) was first reported by Eversole et al. in 1984 and was recently included in the World Health Organization's Classification of Head and Neck Tumors (WHO-2005). Desmoplastic ameloblastoma shows separate clinics radiographic entity as it differs from the other forms of the ameloblastoma in its anatomical, locality morphology and radiographic appearances.<sup>1,2,3</sup>

### Case Report-

A 17 year-old female patient reported with a case of diffuse swelling on left side of mandible extending antero-posteriorly from midline to angle of mandible and superior- inferiorly from corner of mouth to Submandibular region with an asymptomatic swelling that had started 10 months back. Since then, there had been gradual increase in the size of swelling to its present size. Swelling was painless and bleeding was absent. There was no history of trauma. The physical examination revealed facial asymmetry due to swelling on the left side of face which was oval in shape and had a smooth surface. The skin over the swelling appeared normal and was tender on palpation.



(Fig 1) : Diffuse swelling on left side of mandible extending antero-posteriorly from midline to angle of mandible and superior- inferiorly from corner of mouth to Submandibular region.

The intraoral examination disclosed a large mass, approximately 6x5 cm in size, extending from the left central incisor to the left first molar buccally as well as lingually. Buccolingual expansion of the cortical plate of mandible was evident. The swelling was bony hard in consistency, non-fluctuant, nonreducible, noncompressible and nonpulsatile. The teeth in the affected area were not sensitive to percussion and no mobility could be demonstrated. No lymphad-

enopathies or fistulae were present.

### Radiographic examination

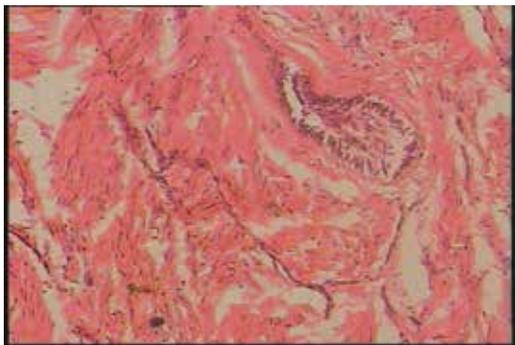
Computed tomography of the lesion showed a predominantly expansile, ill defined lesion extending from left central incisor to mesial root of first mandibular molar having irregular border with mesial resorption of root with an approximate size of 6 × 5 cm. There was loss of lamina dura around the involved teeth. The lesion caused divergence of the root of the lateral incisor without any signs of root resorption.



(Fig 2) : Computed tomography of the lesion showing a predominantly expansile, ill defined lesion extending from left central incisor to mesial root of first mandibular molar

### Histological report

H and E staining shows dense, collagenous, fibrous tissue in which the neoplastic epithelium forms small irregular islands having slender, straggling extensions. There is no cyst formation and ameloblast like cells are present in small foci only at one place. Stellate reticulum like tissue is also absent. Epithelial islands consist of densely packed, spindle shaped cells. Calcification in fibrous stroma and bone is also seen.



(Fig 3): H and E staining shows dense, collagenous, fibrous tissue in which the neoplastic epithelium forms small irregular islands having slender, straggling extensions.

### Discussion

Up until now, less than 150 patients have been reported in the literature. A painless swelling or bony expansion is the most conspicuous clinical manifestation in most of the cases. Desmoplastic ameloblastoma constitutes 0.9% to 12.1% of all ameloblastomas. The mean age at initial presentation is 42.3 years (range 17-70 years). Till 2009 Sixty-two lesions occurred in the mandible and fifty-one lesions in the maxilla. Approximately half of the desmoplastic lesions are located in the maxilla, and the vast majority of them occur in the anterior or premolar portion of the jaws. This is in contrast to the location of the unicystic or classic types of ameloblastoma, which usually are found in the posterior area of the mandible. Maxillary lesions are more insidious than mandibular tumors because of very thin cortical bone forms a weak barrier for the spread of tumors earlier and more quickly than do mandibular neoplasms leading to an early invasion of adjacent structure. The diffuse radiographic appearance and 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. The patient described in this report presented no root resorption, but there was displacement of the adjacent teeth.

Radiologically, the lesion often presented multilocular (49.3%; 36/73), mixed radiolucent/radiopaque (55.6%; 50/90) and with ill-defined borders (64.0%; 48/75) resembling benign fibro-osseous lesions. Histopathologically it always shows extensive stromal desmoplasia with abundance of thick collagen fibers that seem to compress the epithelial islands. The tumor grew very slowly and did not appear to have a strong potential for local extension like typical ameloblastomas.<sup>4, 5, 6</sup>

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.<sup>7</sup>

It is suggested that desmo-plastic 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.<sup>7</sup> Immunohistochemical studies suggest that the desmoplasia originates from de novo synthesis of extracellular matrix proteins. Because ameloblastomas are known to recur years after initial treatment, the need for long-term periodic follow-up to avoid recurrence should be emphasized.<sup>8</sup>

### Conclusion

We conclude on the available information that there is need to recognize the distinction in the observed biologic profile of DA and conventional ameloblastoma, especially because DA may present a confusing clinical diagnosis in view of its resemblance to a fibro osseous lesion. However, further studies are necessary to clarify the biologic profile of DA, especially the induction of desmoplasia observed in this lesion.

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