

## Case Report

### Mucous Cell Differentiation in Desmoplastic Ameloblastoma: Unique Presentation in Posterior Mandible

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#### Abstract

Desmoplastic ameloblastoma is an unusual type of ameloblastoma characterized by pronounced desmoplastic stroma. The occurrences of mucous cells are even rare phenomenon in desmoplastic ameloblastoma and to date only two cases have been reported in the literature in anterior and premolar area. This is a unique case report of desmoplastic ameloblastoma in 25 year old female patient with mucous cell differentiation in posterior mandible as all the previous published cases showed occurrence in anterior jaws. The possible pathogenesis for mucous cell differentiation reflects the pluripotential character of the odontogenic epithelium.

**Keywords:** Odontogenic Tumors;Jaw Tumours;Neoplasms by Histologic Type;Desmoplastic; Ameloblastoma.

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#### Introduction

Ameloblastoma, the most common odontogenic epithelial neoplasm is derived from various sources of odontogenic epithelium. There are various histologic subtypes but ameloblastic differentiation is a histopathological hallmark of ameloblastoma. Various histopathology subtypes of ameloblastoma have been recognized such as follicular, acanthomatous, granular cell, basal cell, plexiform and desmoplastic etc.<sup>1</sup> Desmoplastic ameloblastoma has specific clinical, radiographical and histopathological feature.<sup>2</sup> In addition this tumour predominantly involves the anterior region of the jaw which is an unusual location for ameloblastoma.<sup>3</sup> Further, ciliated or mucous cell metaplasia in odontogenic cyst is well known.<sup>4</sup> The presence of mucous cells in the epithelial linings of ameloblastoma is rare. The purpose of this article is to present additional case of desmoplastic ameloblastoma that has occurred in an unusual site with distinctive histopathological features which makes this case unique along with brief review of literature.<sup>2</sup>

#### Case Report

A 25 year old male reported to the department of oral pathology and microbiology with a chief complaint of painless swelling in the lower left posterior region of the mandible (Figure 1a). The patient had noticed a gradual increase of the swelling for two months before consultation.

Intraoral examination revealed bony swelling in the mucogingival area of the left posterior mandible extending from first premolar to distal surface of first molar without inflammation of overlying mucosa. Both the cortical plates were expanded and tooth #36 and #38 were carious. All teeth were present except #38. Radiographically, an ill-defined tumorous lesion with radiolucency involving the tooth #34, #35, #36 and #37 was evident. In occlusal and panoramic radiograph the lesion was located in the apical region extending from mandible first premolar to distal surface of first molar without root resorption (Figure 1b). Computed tomography image demonstrated osseous protrusion of the mandible associated with the tumorous lesion (Figure 1d). Incisional biopsy was carried out, which showed animal like pattern of odontogenic islands squeezed out in collagenous stroma. Diagnosis of desmoplastic ameloblastoma was confirmed. On the basis of which excisional biopsy was performed in the same institution.

On gross examination segmental resection of the left mandible revealed thick mucinous material from the lesional surface after removing the partially resorbed buccal cortical plate from the specimen (Figure 1c). Multiple tissues were selected from surgical specimen and processed. Sections were stained firstly with hematoxylin and eosin and secondly by mucicarmine stain to confirm the presence of mucin.

Histopathological examination exhibited odontogenic epithelial islands in kite-tail or animal like configuration in dense collagenous connective tissue stroma. The striking feature of the excisional biopsy revealed many odontogenic islands which were dilated, along with few compressed islands in a collagenous stroma. Each island was peripherally lined by tall columnar odontogenic epithelial cells and centrally placed stellate reticulum like cells along with

cystic spaces. These dilated odontogenic islands also showed signet ring shaped mucous cell differentiation closely associated with squamous metaplasia of stellate reticulum like cells, which were mucicarmine positive. Few islands also showed mucous pool (Figure 1e, f, g and h). Inflammatory change was absent in the stroma which was totally purely collagenous. Lesional tissue showed new bone formation as a feature of desmoplastic ameloblastoma.

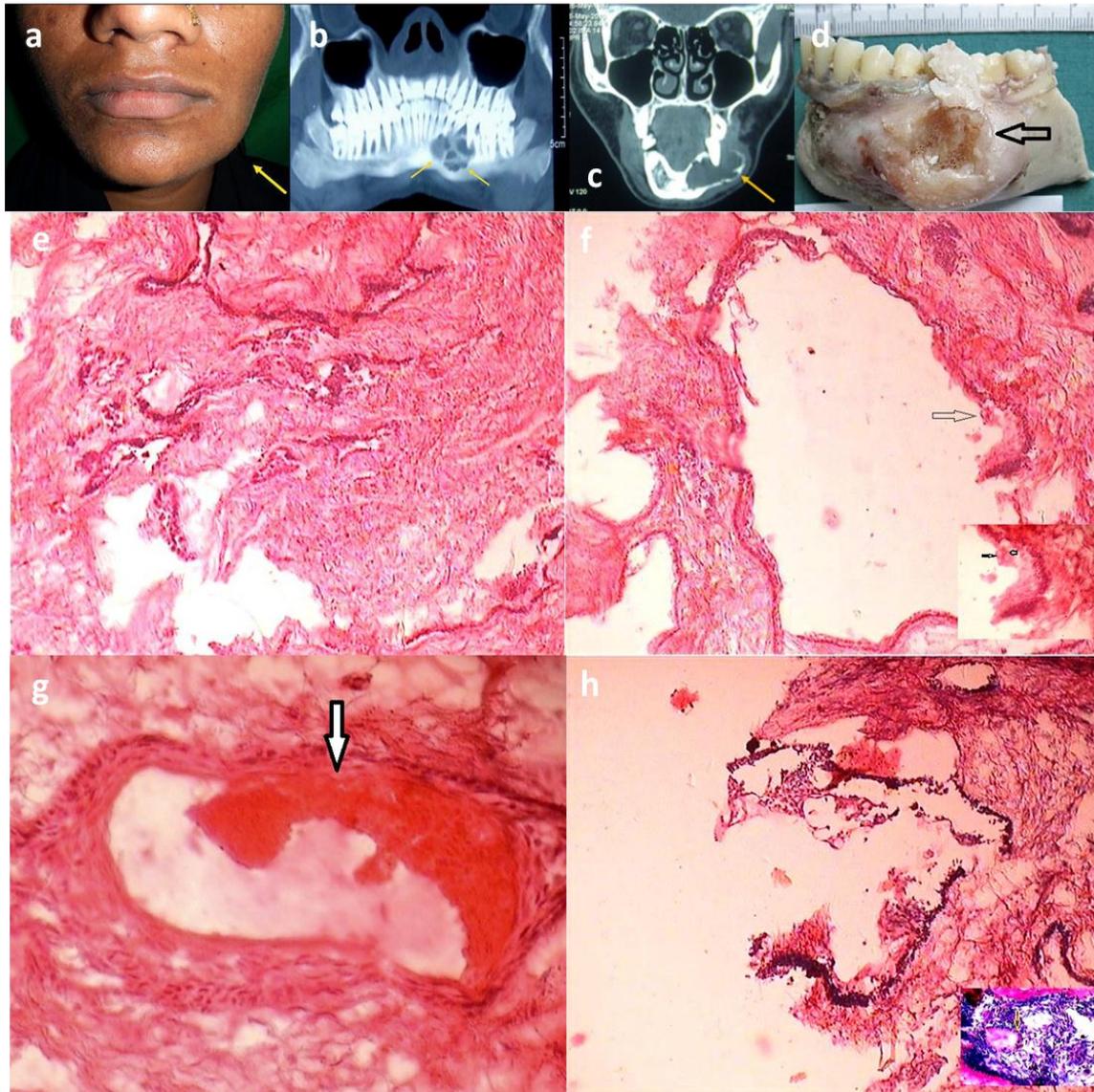


Figure 1: The extraoral photograph showing swelling in the left mandible (a). The radiograph shows a well-defined multilocular radiolucency (b). The excised specimen shows mucinous material in lesional area involving premolar and molar region (c). The CT image shows extensive radiolucent lesion (d). The hematoxylin and eosin stained photomicrograph of the Incisional biopsy shows compressed odontogenic islands in dense collagenous stroma under low-power view (e). The excisional biopsy shows dilated odontogenic epithelial islands in dense collagenous stroma with prominent mucus cells and squamous metaplasia (inset shows mucous cell) (f). The photomicrograph shows mucus pool in center of Odontogenic Island (g), and squamous metaplasia (inset shows mucicarmine positivity for mucous pool) (h).

Author	Age / Sex	Location	Diagnosis	Site of mucous cell observed
Hertenian & Kalfayan	53 / F	Anterior Mandible	Conventional Solid	Solid tumor islands
Van Wyk et al	21 / F	Anterior Maxilla	Unicystic	Cystic structures
Ranbenheimer et al	-----	Anterior Mandible	Conventional Solid	Solid tumor islands
Takata et al	24 / F	Anterior and Left Mandible	Desmoplastic	Cystic structures
Wilson et al	31 / M	Anterior Mandible	Conventional Solid	Solid & Cystic structures
Yoon et al	24 / M	Right Mandible	Unicystic	Cystic structures
Punnya et al & Rekha et al	32 / M 17 / M	Anterior Maxilla Anterior Mandible	Desmoplastic & Follicular (2 cases)	Cystic and Solid Structures
Sandhya Tamgadge et al (Present case)	25 / F	Posterior Mandible	Desmoplastic	Cystic and Solid Structures

Table 1: Review of literature of mucous differentiation in Ameloblastoma.

### Discussion

Desmoplastic ameloblastoma was first described by Eversole et al 1984 as an unusual variant of ameloblastoma which was histologically characterized by desmoplastic stroma.<sup>5</sup> It has been characterized by WHO as a variant of ameloblastoma with specific clinical, imaging and histopathological features.<sup>6</sup>

According to our review, predilection of occurrence was seen in anterior region of either maxilla or mandible as also reported by Kawai *et al*. However our case differed considerably because of its occurrence in the premolar and molar region of mandible along with mucous cell differentiation in desmoplastic ameloblastoma. so far only 2 cases of desmoplastic ameloblastoma with mucous cell differentiation have been reported but in anterior mandible. However our case differed considerably because of its occurrence in the premolar and molar ramus region of mandible.

Histopathologically irregular odontogenic islands with a stretched out kite-tail or animal like configuration in a dense desmoplastic stroma is evident. The most interesting aspect of this lesion was the presence of mucous cell component in a stretched out but dilated odontogenic islands in a dense collagenous stroma, which is rarely reported in the literature with mucous pool in a few. Ameloblastoma rarely include mucous cells. Till date only 8 cases have been reported.<sup>4,7,8</sup> (Table 1). All these cases

occurred in anterior or premolar area of the jaws. Amongst the various histopathological variants of ameloblastoma only two desmoplastic ameloblastoma showed mucous cell differentiation reported by Takata et al in the anterior region of left mandible and Punya et al in anterior mandible. But the present case was present in the premolar and molar region which is a unique finding also odontogenic islands were expanded or dilated except in few areas instead of compressed as mentioned by Riechart.<sup>6</sup> This case also showed mucous cells with cystic structures as reported by Takata et al. Predominantly mucous cell were associated with squamous metaplasia, suggesting a close relation between two cell types as reported by Punya et al.<sup>8</sup> The possible pathogenic mechanism for above squamous and mucous cell would appear to be a reflection of the pluripotential character of the odontogenic epithelium.<sup>6</sup>

### Conclusion

As occurrence of mucous cell in ameloblastoma is not a rare finding but desmoplastic ameloblastoma with mucous cell differentiation in the premolar and molar area is very rare as per literature. The prognosis of desmoplastic ameloblastoma with mucous cell differentiation cannot be predicted as only few cases have been reported in the literature. Desmoplastic ameloblastoma can show expanded and dilated, stretched out odontogenic islands in collagenous stroma in contrast to compressed odontogenic islands. It can

affect the premolar and molar area depending upon the chronicity of the lesion.

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