

## Case Report

## Hybrid Ameloblastoma - An Unusual Combination of Desmoplastic and Conventional Ameloblastoma.

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**Abstract:** Ameloblastomas are benign, locally aggressive neoplasms arising from the odontogenic epithelium. The follicular type is of the most common variant while desmoplastic type is amongst the uncommon variant of ameloblastoma. Radiologically, the former appears as unilocular or multilocular lucency, while the latter usually appears as mixed radiopaque-radiolucent lesion. When the desmoplastic type coexists with other common variants; it is termed as hybrid ameloblastoma. A case of hybrid ameloblastoma, which was a combination of desmoplastic and follicular types is being reported because of its rare occurrence and unique radiological picture.

**Keyword:** Desmoplastic, follicular, ameloblastoma, hybrid

### INTRODUCTION

Ameloblastomas are the commonest benign but locally aggressive neoplasms of proliferating odontogenic epithelial origin<sup>1,2</sup>. Among the multiple histological types, follicular and plexiform patterns are the most common; followed by acanthomatous and granular cell types. Uncommon variants include desmoplastic, basal cell, clear cell, keratoameloblastoma and papilliferous keratoameloblastoma. 'Hybrid' lesion of ameloblastoma is composed of desmoplastic ameloblastoma and conventional follicular/ plexiform ameloblastoma. This is an unusual variant of ameloblastoma with only 8 cases so far reported in the literature<sup>3</sup>. Desmoplastic ameloblastoma exhibits important differences compared with the classic type. Hybrid lesion will show the features of both conventional and desmoplastic variant, making management difficult a thorough preoperative radiological evaluation is important. The latter not only helps in diagnosis but also serves as a guide for the site of biopsy and delineates the extent of tumor.

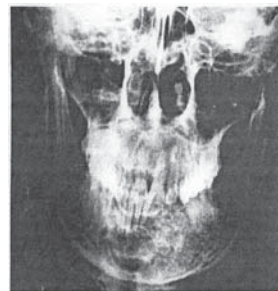
### CASE REPORT

A 34-years female came to us for radiological evaluation with, painless hard swelling of in the lower jaw of 12-15 month duration. Clinical examination revealed bony hard swelling arising from the anterior lower jaw. The intraoral examination revealed a nontender firm to hard mass in the region of anterior mandible, covered by a red but intact and mobile mucosa. The lesion extended from the premolar region on the left side to the premolar region on the right side. No lymphadenopathy or fistulae were present. The involved teeth were vital but were displaced lingually. Past medical history was unremarkable.

The posteroanterior radiograph of the mandible revealed a diffuse, solid, illdefined, predominantly radiopaque lesion interspersed with fine radiolucent areas producing a *honeycomb appearance* on left side with a cystic component on the right side with an apparent *floating tooth* appearance of the anterior incisors and canine on right side (fig 1).

The noncontrast computed tomography revealed the nature of inter matrix of the lesion along with its extent. There was solid, expansile, poorly defined mass with a matrix of predominantly radiodense with intervening radiolucent areas resembling honeycomb in the anterior part of mandible extending from the incisor to premolar region on left side. A unilocular cystic component was also observed the lamina dura of the teeth in the affected mandible along with destruction of both buccal and lingual cortices and a soft tissue bulge both intraorally and extraorally. (fig.2). There was complete loss of corticomedullary differentiation. Based on the radiological

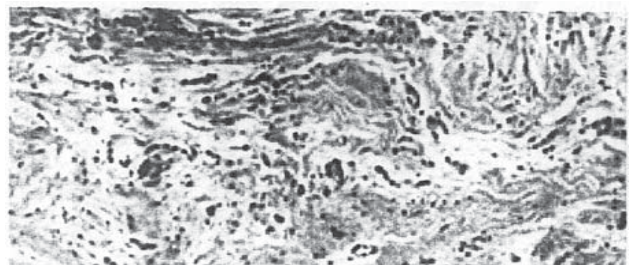
appearances, a provisional diagnosis of desmoplastic variant of ameloblastoma with concomitant follicular / plexiform variant was made. Incision biopsy from solid and cystic areas of the tumor was done which confirmed the concomitant desmoplastic and follicular variant of ameloblastoma (fig.3). The patients then underwent hemimandibulectomy across the midline with prosthesis; postoperative period was uneventful.



**Figure 1:** Radiograph of mandible (PA view) Shows a diffuse radiodense lesion with honeycomb appearance in the anterior mandible on left side and a radiolucent cystic lesion on right side of midline with lingual displacement of the adjacent teeth



**Figure 2:** Axial CT image shows an expansile, solid, mixed radiodense radiolucent lesion with poorly defined margins on the left side contiguous with a cystic component on right side occupying the anterior mandible associated with destruction of the cortices and extension into the soft tissues



**Figure 3:** Photomicrograph showing features of both desmoplastic and follicular ameloblastoma

### DISCUSSION

The desmoplastic ameloblastoma is an unusual variant of ameloblastoma characterized by low occurrence rate and marked stromal desmoplasia.

These tumors are most commonly seen in 3<sup>rd</sup> to 5<sup>th</sup> decade of life with a male preponderance. These lesions occur in maxilla and mandible with nearly the same frequency in the anterior or premolar portion of the jaw. In contrast, classic ameloblastoma occurs primarily in the posterior areas of mandible. Maxillary lesions are considered more complex as they have easy access to the adjacent vital structures and the maxillary sinus. Besides this, the very thin cortical bone of the maxilla forms a weak barrier for the spread of these tumors<sup>4</sup>.

On radiograph and CT, desmoplastic ameloblastoma is characterised by mixed radiolucent & radiopaque appearance often described as honeycomb appearance. It presents a multifocal appearance of minute or punctate osseous flecks because of the infiltrative growth pattern of tumor into surrounding bone with simultaneous vigorous osteoblastic activity<sup>5</sup>. Radiologically, the lesion mimics benign fibro-osseous lesions. Rarely, the tumor may be unilocular or multilocular and may occur in the molar region of mandible<sup>6</sup>. These lesions, however exhibit very high incidence of poorly defined borders, which is in contrast to usual fibroosseous lesion that usually have well defined margins. Computed tomography delineates the internal structure and extent of the lesion accurately and is particularly helpful in determining the border of the lesion and invasion in to the adjacent structures<sup>7</sup>.

MR Imaging may show heterogeneous hypo to intermediate signal intensity on T1-weighted images and heterogeneous hyperintensity on T2 weighted images. Heterogeneous and moderate enhancement may be observed on post-gadolinium T1-weighted images<sup>8</sup>. Histopathology of desmoplastic ameloblastoma reveals small cords of odontogenic epithelium separated by dense, fibrous, connective tissue. Spicules of mature lamellar bone trabeculae have been reported in intimate contact with the tumor, and invasion has been demonstrated<sup>9</sup>.

Desmoplastic ameloblastoma is more aggressive than its other variants. This is suggested by its potential to reach large size, production an early invasion of adjacent structures, diffuse & ill-defined radiographic appearance and the histological presence of bone invasion<sup>9</sup>. Recent literature describes the occurrence of hybrid ameloblastoma with histological features of follicular / plexiform ameloblastoma with features

of desmoplastic ameloblastoma<sup>10</sup>. Radiologically, it may show feature of desmoplastic and conventional ameloblastoma i.e. mixed radiodense-radiolucent lesion along with purely cystic component<sup>11</sup> as was seen in our case.

Thus, since the nature of the lesion, its interface with normal bone and its extent is significant for optimal management, preoperative radiological evaluation of lesion in the anterior jaw is very essential. Fibro-osseous appearing lesion may be characteristic of desmoplastic ameloblastoma on CT. CT may also delineate the concomitant presence of cystic component of the mass that should arouse the suspicion of hybrid ameloblastoma. A thorough radiological examination will help making a preoperative diagnosis, guiding the site for biopsy and thus reaching a definitive diagnosis preoperatively, preventing errors in formulating treatment plans and achieving higher cure rates in such lesions.

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