

# A combination of follicular and plexiform ameloblastoma: A rare case report

### ABSTRACT

Ameloblastoma is a benign but locally aggressive neoplasm of odontogenic epithelium origin. Follicular type is the most common variant of ameloblastoma followed by plexiform, acanthomatous, and granular subtypes. Synchronized existence of plexiform ameloblastoma with follicular ameloblastoma is a rare event. When the plexiform type coexists with other common variant, it is termed as hybrid ameloblastoma. Here, we present a case of hybrid ameloblastoma with follicular and plexiform variants occurring in a 43-year-old female patient having persistent swelling for 5–6 months in the left mandibular body region extending up to the left angle of the mandible. The tumor was treated by en bloc resection followed by reconstruction.

**Keywords:** Ameloblastoma, follicular, odontogenic neoplasm, plexiform

### INTRODUCTION

Ameloblastoma is the most common benign but locally aggressive neoplasm of proliferating odontogenic epithelial origin. It is a challenge to pathologists because of its diversity of histological features and to surgeons due to its frequent defiance to complete eradication. It accounts for approximately 1% of all tumors and cysts that occur in the jaw and can develop at any age, with a peak prevalence in the third and fourth decade.

Ameloblastomas can occur at any location in the mandible or maxilla, but the regions of the mandibular molars and ramus are the most prevalent anatomical locations (80%). When the tumor occurs in the maxilla, the posterior region is most commonly involved.<sup>[1]</sup>

Radiographically, ameloblastoma may represent as unilocular or multilocular radiolucent image in the shape of “soap bubble” or “honeycomb.” Histopathologically, ameloblastoma exhibit proliferating odontogenic epithelium within a background of fibrous stroma.

The chief histopathological variants of ameloblastoma are the follicular and plexiform types, followed by the acanthomatous and granular cell types. Uncommon variants

include desmoplastic, basal cell, clear-cell ameloblastoma, keratoameloblastoma, and papilliferous ameloblastoma.<sup>[2]</sup>

A possible transient form of plexiform ameloblastoma showing microscopic features of plexiform variant together with areas of classical follicular ameloblastoma has been described as hybrid ameloblastoma.

The report which we are presenting is a case of hybrid ameloblastoma in which there is histologically dual picture of follicular and plexiform ameloblastoma.

### CASE REPORT


A 43-year-old female patient reported to our department with

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the chief complaint of swelling in the left lower jaw region for 5–6 months. A history of progressive increase in size was elicited. There was no history of any pain, pus discharge, paresthesia, or anesthesia in the affected jaw. Past dental history includes extraction of multiple upper and lower teeth due to periodontal problem 1 year ago. Past medical history was noncontributory.

Extraoral examination revealed an irregular swelling in relation to the left body of the mandible extending up to the angle region of mandible and also involving the lower border of the mandible. The overlying skin was normal in appearance. The swelling was nontender, firm, nonfluctuant, noncompressible, and fixed to underlying structures. Submandibular lymph nodes of the affected side were nonpalpable. Intraoral examination revealed a single bony hard swelling measuring about 6 cm × 4 cm in size with ill-defined margins, extending from the left lower lateral incisor to the lower left angle region and also involving the left buccal vestibule causing its complete obliteration. The overlying mucosa was normal in appearance [Figure 1a and b].

Orthopantomogram revealed a well-defined, multilocular radiolucent lesion in the left body of the mandible with flecks of radiopacity extending from mandibular left lateral incisor to clinical angle region of the left side. The lower border of the mandible was also involved [Figure 2].

An incisional biopsy under local anesthesia was done, and the specimen was sent for histopathologic examination. Histopathological report of specimen suggested ameloblastoma of follicular type only.

Based on the clinical, radiographical, and histopathological examination, segmental resection of the mandible was planned. Under complete aseptic condition, segmental resection of the mandible was done under general anesthesia with respect to the left lateral incisor up to the left mandibular angle region. Bony defect was reconstructed with locking reconstruction plate to give reasonable cosmetic and functional outcome to the patient. Layer-wise closure was done, and pressure dressing was given [Figure 3]. Whole resected segment was sent for histopathologic examination.

#### Histopathologic examination

Histopathological examination revealed long anastomosing cords or larger sheets of epithelium which were bounded by columnar and cuboidal ameloblast like cells surrounding more loosely arranged epithelial cells. Supporting stroma tends to be loosely arranged and vascular.

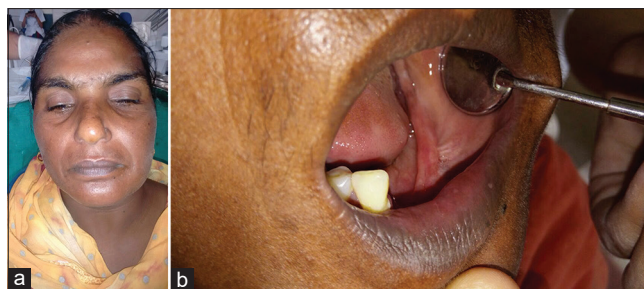


Figure 1: (a) Preoperative extraoral image. (b) Preoperative intraoral image

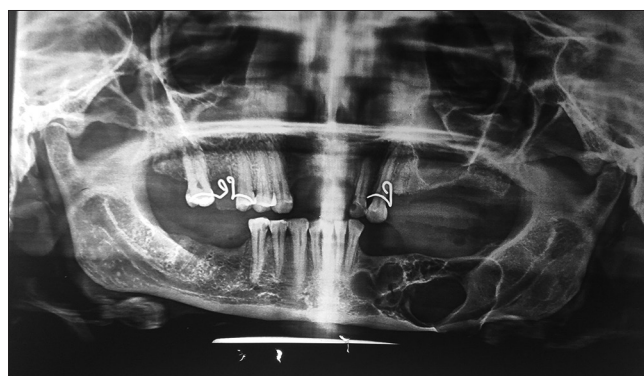


Figure 2: Preoperative orthopantomogram

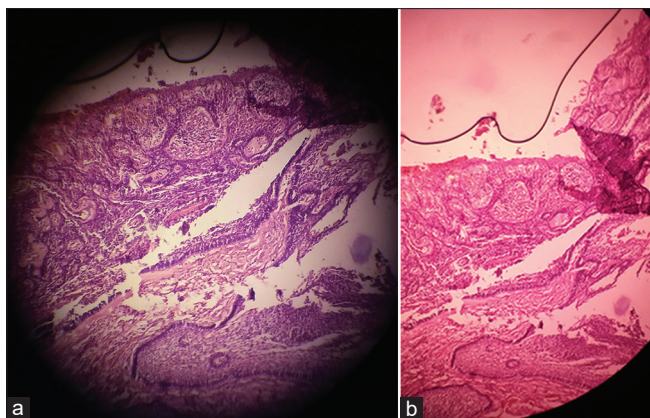


Figure 3: Postoperative orthopantomogram with reconstruction plate

It was also composed of many small discrete islands of tumor composed of a peripheral layer of cuboidal or columnar cells whose nuclei were generally well polarized. This closely resembles ameloblasts and preameloblasts. The central part of follicle was showing stellate reticulum like cells. Histopathological findings were suggestive of both plexiform and follicular ameloblastoma [Figure 4a and b].

#### DISCUSSION

Ameloblastomas account for 1% of all tumors of the jaw encountered during the third to fifth decades of life. About 80% of all cases occur in the mandible, of which 70% cases are seen in the ramus. Follicular and plexiform are the commonly encountered variants accounting to 32.5% and 28.2%, respectively, followed by the acanthomatous subtype 12.1%,



**Figure 4: (a) Histological appearance of hybrid ameloblastoma as low power. (b) Histological appearance of hybrid ameloblastoma at high power.**

while desmoplastic is extremely uncommon with incidence rates ranging from 4% to 13%.<sup>[2,3]</sup>

Histopathologically, hybrid ameloblastoma shows areas of compressed odontogenic epithelial islands along with areas of typical plexiform/acanthomatous/desmoplastic ameloblastoma.<sup>[4,5]</sup> Some cases of hybrid ameloblastoma also showed granular cell transformation in some of the tumor cells along with the areas of follicular and plexiform ameloblastoma while some showed basaloid changes also. As the present case gives a mixed picture of plexiform and follicular ameloblastoma, it can be considered as a case of hybrid ameloblastoma histopathologically.<sup>[6,7]</sup>

Whether the hybrid ameloblastoma should be considered as a hybrid tumor or collision tumor is a matter of debate since its existence. While collision tumors are considered as two lesions arising from independent topographic sites, a tumor is considered to be hybrid if two or more disparate and well-established tumors exhibit obvious differentiation.<sup>[8]</sup> The hybrid variant of plexiform ameloblastoma shares common clinical characteristic features with typical ameloblastoma, i.e., no definite gender prevalence, site predilection of the posterior region of jaws, and mixed radiolucent-radiopaque appearance, except for the histologic presence of areas of conventional ameloblastoma along with areas of plexiform ameloblastoma with a striking mandibular predilection pointing toward differentiation concept.<sup>[9]</sup>

In the present case, 43-year-old female patient reported in our department with persistent painless slow-growing swelling involving the left body of the mandible region for the past 5–6 months. There was no history of any pain, pus discharge, paresthesia, or anesthesia in the affected jaw.

Histopathological features revealed both follicular as well as plexiform ameloblastoma which adds to unique case till

reported in literature. Ameloblastoma was known for its high recurrence rate if excision was incomplete and same is true with hybrid ameloblastoma. Therefore, the treatment of choice was surgical excision with wide-free margins.

Bachmann and Linfesty reported a solid/multicystic type ameloblastoma that showed a variety of histologic types, with plexiform and follicular predominating and was treated with partial resection of the mandible.<sup>[10]</sup>

The present case was treated with segmental resection followed by reconstruction of deformity with reconstruction plate fixed with locking screws under general anesthesia. She made uneventful recovery during her follow-up visits and is still under follow-up.

The challenge in the management of large ameloblastoma of the mandible is not only to excise the tumor completely to prevent recurrence but also to provide reasonable cosmetic and functional outcome to the patient.

## CONCLUSION

Ameloblastomas are an enigmatic group of oral tumors. Ameloblastomas not only occur in the posterior region but can also occur in the anterior region of the jaw. Since variants of ameloblastoma differ in biologic behavior, the histopathological examination is essential along with clinical examination which is of significance to the clinician for effective treatment plan as well as to prevent recurrence.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Nil.

## Conflicts of interest

There are no conflicts of interest.

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