



Case Report

# Malignant Odontogenic tumor: a case report

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## ABSTRACT

Malignant odontogenic tumors are rare and represent approximately 1% of all oral malignancies. Ameloblastic Carcinoma is an uncommon malignant odontogenic tumor accounting for 1.5%-2.0% of all odontogenic tumors that can be difficult to differentiate from Ameloblastoma, and can arise directly as an undifferentiated lesion or from a preexisting benign lesion. It is a highly aggressive tumor with rapid growth and a high potential for distant metastasis. The most common site of occurrence is posterior mandible. Here we present a case report of Ameloblastic Carcinoma, relapse case of a benign lesion 15 years after surgery which presented as firm growth in the right mandible for 2 months.

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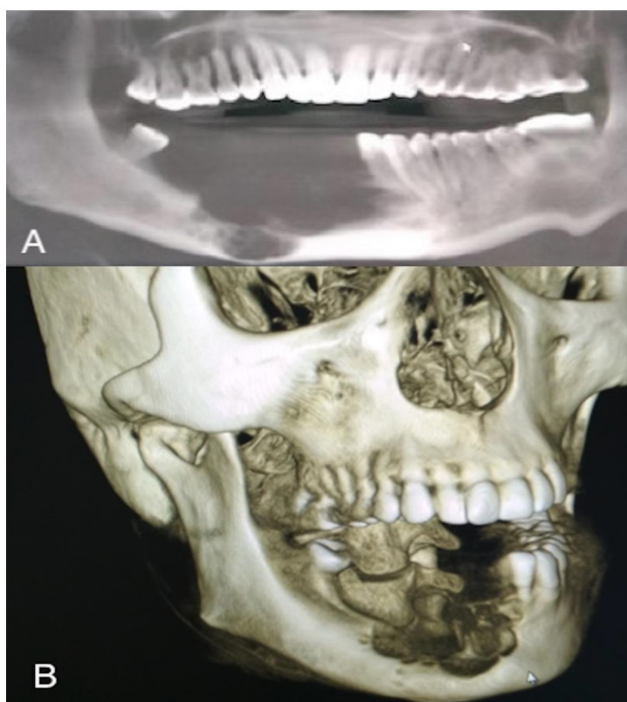
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## INTRODUCTION

Ameloblastic carcinoma is rare, malignant, odontogenic neoplasm that histologically has retained the features of ameloblastic differentiation and also exhibits cytological features of malignancy. The term Ameloblastic carcinoma (AC) was introduced by Shafer in 1974.<sup>1</sup> Ameloblastic Carcinoma has wide age range with male predilection and posterior mandible being the commonest site.<sup>2</sup> The majority of ameloblastic carcinomas arise de novo, ex-ameloblastoma or ex-odontogenic cyst. Lesion is symptomatic with swelling and pain which shows paresthesia of affected region. Radiographically this lesion shows multilocular radiolucency mimicking solid multicystic ameloblastoma.<sup>1</sup> Herein we report a case of ameloblastic carcinoma in a 36 years old male patient.

## CASE REPORT

A 36-years-old male patient came to the OPD with chief complain of swelling in the lower right region of the

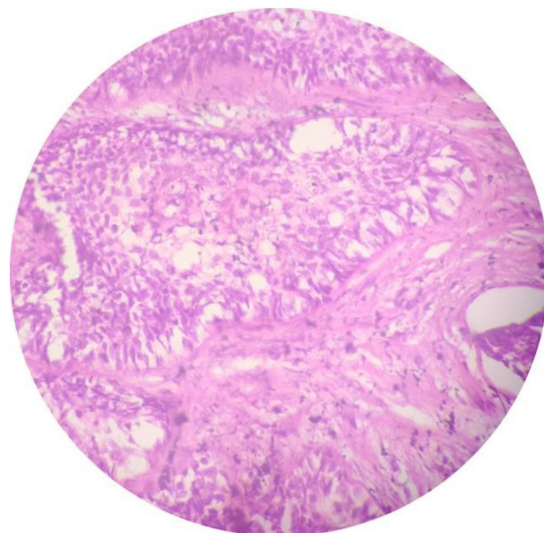


**Figure 1: A) CBCT showing multilocular radiolucency from mesial of 32 to distal of 44. B) 3D reconstruction showing perforation of buccal and lingual cortical plates and thinning of the lower border of mandible**

mandible for 2 months. Swelling was non-tender and was gradually increasing in size. The patient claimed to have a surgery done 15 years back on the same region for previous history of ameloblastoma. There was no gross facial asymmetry with palpable, stony hard right sub mandibular and sub mental lymphnodes detected on extra oral examination. Similarly, intra oral examination revealed missing 31, 41, 42, 43, 44, 45 and 46 with expansion of the buccal and the lingual cortices in relation to 31, 41, 42 and 43. The over lying mucosa was normal.

On radiographic examination, cone beam computed tomography (CBCT) revealed multilocular radiolucency from mesial of 32 to distal of 44 (fig. 1A). Perforation of buccal and lingual cortical plates with the thinning of the lower border of mandible could also be appreciated. The surrounding bone had normal trabecular pattern (fig 1B). On the basis of clinical and radiographic findings provisional diagnosis of ameloblastoma of right body of mandible was given.

The incisional biopsy was received and sent for histopathological examination which revealed nests and islands of tumor cells having ameloblastomatous origin. Central cells showing atypical features with increased mitotic figures, squamous metaplasia and keratin pearl could be appreciated (fig. 2). Based on these findings histopathological differential diagnosis of Ameloblastic carcinoma and aggressive Acanthomatous ameloblastoma was given. Hemi mandibulectomy with radial neck



**Figure 2: Showing nest and island of tumor cells ameloblastomatous in origin, central cells showing atypical features and increased mitotic figures (HE stain, X400)**

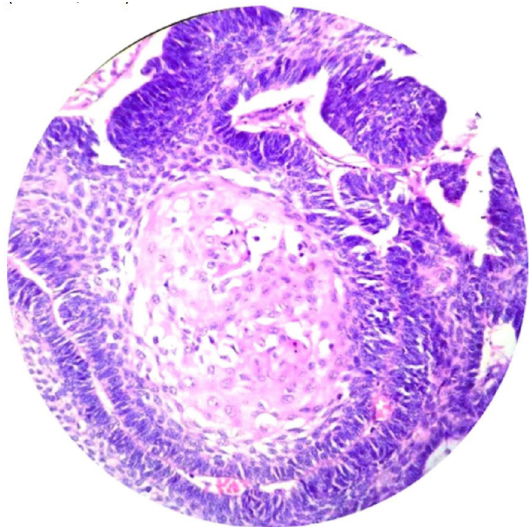
dissection upto level III and reconstruction was done as treatment. Resected mandible of about (9.5x 2.5x 3)cm along with the lymphnodes were received. Soft tissue from buccal and lingual side and the total of 12 lymphnodes were sent for histopathological examination. A piece of bone was also sent for decalcification.

Microscopic examination revealed nest and islands of ameloblastomatous odontogenic epithelium within the stroma of mature fibrous tissue. Nests and islands show peripheral palisading dark columnar cells with reverse nuclear polarization, central area containing loosely arranged cells resembling the stellate reticulum of enamel organ. Mild to moderate cellular pleomorphism, keratin pearl formation could also be appreciated with frequent mitotic figures present up to 3-5/hpf. Lymphnodes and bone were free of tumor cell invasion (fig. 3). Thus, based on clinical, radiological and histopathological findings final diagnosis of Ameloblastic carcinoma was given.

## DISCUSSION

Ameloblastic carcinoma is neoplasm in which the histologic pattern of a solid multicystic ameloblastoma has been retained in the primary growth in the jaws and/or in any metastatic growth, yet also exhibits cytological features of malignancy.<sup>1</sup> It is uncommon malignant lesion accounting for 1.5%-2.0% of all odontogenic tumors.<sup>3</sup>

It is sub classified as primary type and secondary type. Primary type: Histologically defined as an apparent malignant neoplasm that exhibits cytological malignancy, including pleomorphism, hyperchromatic nuclei, and high mitotic rates, and has the histologic features of ameloblastoma without pre-existing history. Secondary



**Figure 3: Showing nest and islands of ameloblastomatous odontogenic epithelium within the stroma of mature fibrous tissue with peripheral palisading dark columnar cells, Mild to moderate cellular pleomorphism, keratin pearl formation, Mitotic figures frequently present (3-5/hpf) (H&E stain, X400)**

type: It is thought to occur from the recurrent or pre-existing ameloblastoma having the histologic features of a coexisting ameloblastoma. Ameloblastic carcinoma, secondary type, is extremely rare<sup>4</sup> which was seen in our case.

This lesion frequently occurs between the age of 15 to 84 years. It occurs most commonly in male with the male: female ratio 1.4:1. The commonest site of occurrence in posterior mandible.<sup>5</sup> Radiographically it may resemble solid multicystic ameloblastoma. In most cases, they present as ill-defined radiolucencies. Foci of radiopacities, probably due to dystrophic calcification, have also been observed.<sup>4</sup> Often lesions present with perforation of the cortical bone which was consistent with our case. Sometimes it may also extend into the neighboring soft tissue.<sup>6</sup>

Histologically, tumor is composed of islands and cords of ameloblastomatous odontogenic epithelium in an infiltrative pattern within a stroma of mature fibrous tissue. The epithelium may reveal a single outer layer of ameloblastic cells of columnar to cuboidal shape which exhibit a tendency for palisading and reverse nuclear polarization.<sup>1</sup> The stellate reticulum within epithelial islands is often condensed and hypercellular, presenting a less orderly pattern.<sup>7</sup> Characteristic differentiating features are nuclear enlargement with granular stippled nucleoplasm, nuclear hyperchromatism, mild to moderate pleomorphism, an increased nuclear cytoplasmic ratio, and increased mitotic activity with abnormal forms of mitoses. Mitotic figures may attain a count of 2 to 5 per high-power field.<sup>8</sup> Individual cell keratinization and keratin pearl formation may be seen. In our case, mild to moderate pleomorphism, keratin pearl formation and increased mitotic figures were important features in establishing final diagnosis.

On immunohistochemistry, lesions show increased expression of CK5 and CK18 moderate to strong staining for AE1/AE3, CK14 and CK19 focal positivity for CK8 and negative for carcinoembryonic antigen (CEA). Alpha Smooth muscle actin (SMA) is supporting tool to distinguish Ameloblastic carcinoma from Ameloblastoma because it is present only in neoplastic cells of Ameloblastic carcinoma.<sup>5</sup>

As far as treatment is concerned, wide local excision is the treatment of choice as most authors have recommended.<sup>9,10</sup> Neither chemotherapy nor radio therapy has shown significant benefit, but they need to be contemplated in advanced cases and for metastatic lesions not amenable to surgical resection.<sup>6</sup> Meticulous follow-up is essential because recurrence and metastasis in the lung and regional lymph nodes have been reported.<sup>6</sup>

## CONCLUSIONS

The management of the colloid cyst needs a multidisciplinary Ameloblastic Carcinoma is a rare entity. Thorough histopathological analysis is obligatory to reach to the final diagnosis. Wide resection and regular follow up is mandatory due to the high chance of recurrence. Study of new cases may further decipher its biological, clinical and histopathological nature.

**Conflict of Interest:** None

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