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Case Report

A rare case report of ameloblastic carcinoma maxilla

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ABSTRACT:
Ameloblast carcinoma is a rare tumor of the facial bones with features of ameloblastoma and carcinoma. It occurs more frequently in the mandible than in the maxilla. Surgical resection with 2–3 cm bone margins, is the current modality for the treatment of AC. Postoperative Radiotherapy may be beneficial who have locally recurred or those with residual disease after resection. The prognosis of AC is poor. The recurrent rate was 50%–90%, whereas the metastatic rate has been reported about 30%.
Keywords: Ameloblastic carcinoma, Odontogenic tumor, Maxilla, Rare and aggressive.

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INTRODUCTION

Ameloblast carcinoma is a rare tumor of the facial bon es with features of ameloblastoma and carcinoma. It occurs more frequently in the mandible than in the maxilla. Ameloblastic carcinoma is an extremely uncommon malignant tumor of odontogenic epithelium accounting for 2% of odontogenic tumor [1]. These tumors have cytologic atypia, poor differentiation and high mitotic index. Usually pesent as a cystic lesion with clinical feature of benign lesion or as a large ulcerated tissue mass with significant bone resorption and tooth mobility[2]. Ameloblast carcinoma occurs mainly in the posterior part of the jaw and has two main types: the first type i s called de novo carcinoma, while the second type is d efined as the malignant transformation of a pre-existing benign ameloblastoma [3, 4].

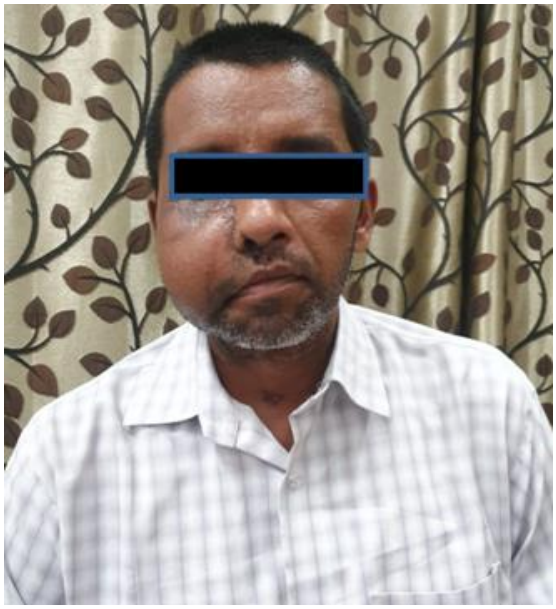
More than 3600 cases of ameloblastomas have been described in the literature [5] but less than 60 cases of ameloblastic carcinoma have been reported so for, among which two thirds occurred in the mandible [6]. Surgical resection with 2–3 cm bone margins, is the current modality for the treatment of AC. Postoperative Radiotherapy may be beneficial who have locally recurred or those with residual disease after resection.The prognosis of AC is poor. The recurrent rate was 50%–90%, whereas the metastatic rate has been reported about 30% [7].

CASE REPORT

A 42 year old male presented with dull aching pain in right upper alveolus the patient underwent tooth extraction in December 2022 at a dental clinic. He noticed a soft tissue mass that had been growing since two to three months. Besides swelling and local site pain the patient did not complaint of any other symptoms including paresthesia or radiating pain. Extra orally the patient showed facial asymmetry due to right maxillary swelling. The skin over the swelling was smooth and normal.

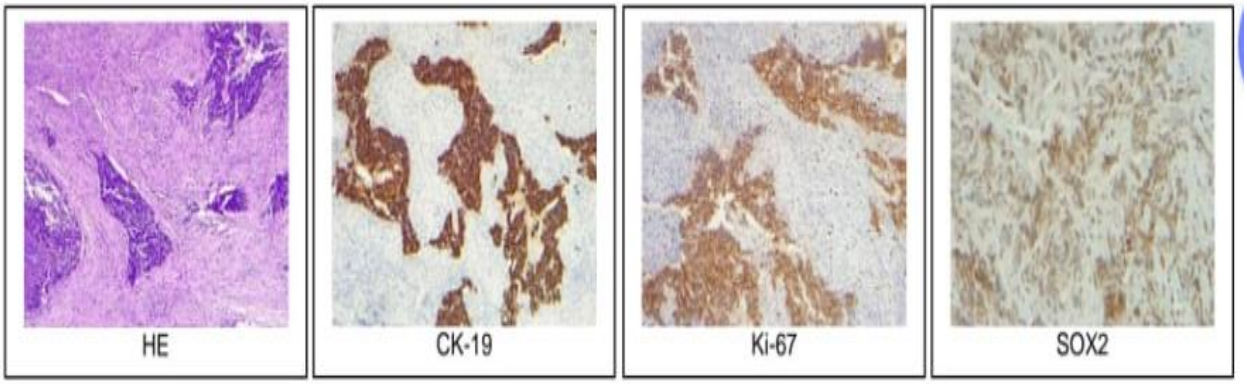
Right upper alveolus cyst specimen was suggestive of epithelial odontogenic neoplasm with the possibilities of sclerosing odontogenic carcinoma, metastatic carcinoma or desmoplastic ameloblastoma with IHC correlation.

The patient underwent Right Radical Maxilectomy + Right Orbital Enucleation +Right Modified Radical Neck Dissection + ALT (left thigh lateral pedicle) Flap in June 2023 at local private hospital (Figure A).



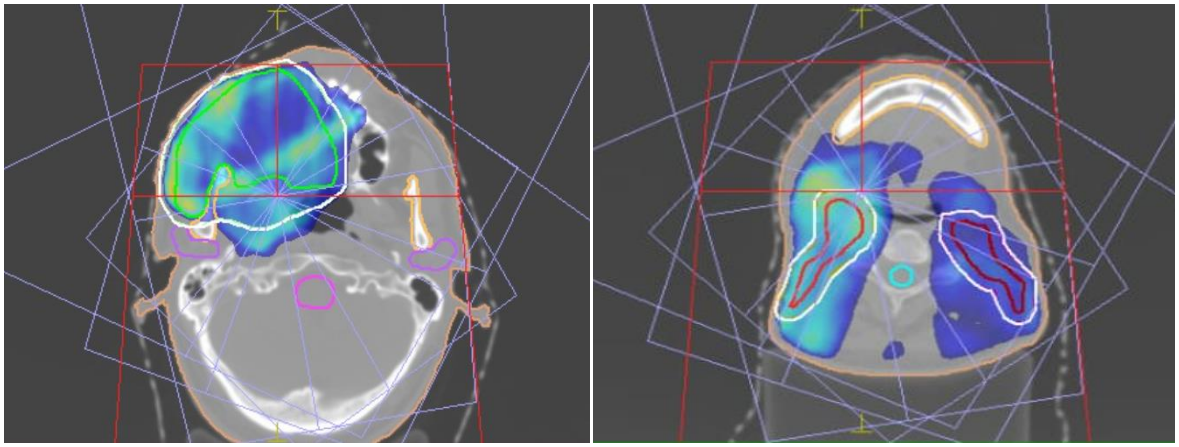
A

(Figure A: shows patient after post operative)



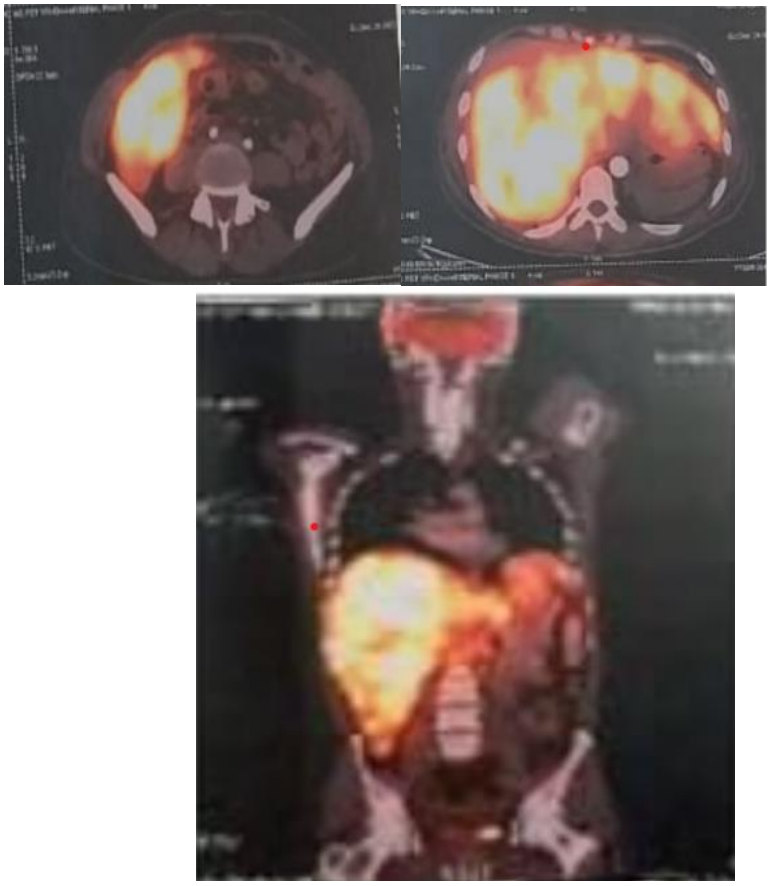
B

(Figure B: shows IHC was suggestive of ameloblastic carcinoma)



C **D**

(Figure C and D: shows IMRT planning)



E

(Figure E showed PET- CT suggestive of distant metastasis)

The histopathological examination report was suggestive of poorly differentiated malignant round cell tumor with lympho-vascular invasion and peri-neural invasion with tumor invading orbital floor. The eyeball was spared by tumor.

The immunohistochemistry (Figure B) was suggestive of ameloblastic carcinoma.

One month after surgery the patient presented to us in cancer O.P.D. initial workup and assessment of the patient for present disease status was done. The patient was taken up for concurrent chemo radiotherapy. The technique employed was Intensity Modulated Radiotherapy (Figure C and D) with a total radiation dose of 66 Gy/30# to PTV and 60 Gy/ 30# to ipsilateral lymph nodes and 54 Gy/30# to contralateral lymph nodes with 6 cycles of concomitant injection cisplatin 50 mg weekly. The patient was kept on regular follow up during the course of entire chemo – radiotherapy.

The patient was kept on regular follow up and CECT neck was done after six weeks of completion of radical treatment. No recurrent lesion was noted for 3 months. Patient complaint of pain abdomen for that ultra-sonography was done which showed space occupying lesions. PET-CT (Figure E) done dated 29/12/23 suggestive of hepatic ,skeletal metastasis with abdominal lymphadenpathy. There was no local recurrence.

CASE DISCUSSION

Ameloblastoma is a common odontogenic epithelial tumor that is benign that can transform into a malignant tumor called ameloblastic carcinoma (AC), which is very rare [8].

Ameloblast carcinoma affects different age groups. Th ere is no genderpreference. The back of the mandible i s the most commonly affected area [9]. Involvement of the maxilla by ameloblastic carcinoma seems to be less frequent than that of the mandible [9]. Swelling, although others include associated pain, rapid growth, trismus and dysphonia is the most common clinical feature [9].

Plain X-ray and computerized axial tomography are radiological investigations.

The osteolytic process may appear on radiographs as a unilocular or multilocular appearance. Metastatic dise ase should be checked, especially in patients with clas sical ameloblastoma, malignant ameloblastoma and a meloblastoma recurrence [10]

Immunostaining distinguish between benign and mali gnant tumors [11]. Other authors have noted, p53 exp ression in AC compared to AM plays a role in malign ant transformation [12].

Treatment of choice is surgical resection. En bloc removal surgical modality with 1–2 cm of normal bone margin to ensure disease-free survival. Local recurrence rates of less than 15% [10].

Ameloblastic carcinoma is considered to be a radioresistant tumor. However, pre- or postoperative radiotherapy may reduce the size of ameloblastic carcinomas [13]. 50% of postoperative patients developed local recurrence or metastasis, and could be treated with radiotherapy. Radiotherapy alone is appropriate for patients who are not surgical candidates, or exhibit advanced local or metastatic disease reported by Dhir K et.al [14].

Local recurrence 0.5–11 years after definitive therapy [15]. Distant metastasis having poor prognosis and may appear as early as 4 months or as late as 12 years postoperatively [15]. Lung is the most common site for a distant metastasis, followed by bone, liver, and brain [15]. Distant metastasis can occur in the absence of a local or regional recurrence as seen in our case [16]

CONCLUSION

In conclusion, ameloblastic carcinoma is a relatively rare type of tumor. Ameloblastic carcinoma exhibit an aggressive clinical behavior, including rapid tumor growth, painful swelling and perforation of the cortex. The proposed mechanisms underlying the transformation of a classic benign ameloblastoma into a malignant tumor remain controversial. It has been indicated that wide local excision with postoperative radiation therapy should be employed. However, novel therapeutic regimens must be considered, including carbon ion therapy and Gamma Knife stereotactic radiosurgery. Controlled studies with larger groups of patients are required to increase the accuracy of results.

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