Ameloblastic Carcinoma of Maxilla and its Management: A Case Report and Review of Literature

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ABSTRACT

The term ameloblastic carcinoma is defined by ameloblastoma with histological evidence of malignancy. Ameloblastoma or ameloblastic carcinoma of the maxilla is a rare entity. We reported a rare case of ameloblastic carcinoma of the left side of maxilla in a 26 year-old male. The patient was treated with surgery followed by adjuvant radiotherapy. The patient was disease free since last 2 years of follow up.

KEYWORDS: Ameloblastic Carcinoma, Maxilla, Management.

INTRODUCTION

Ameloblastoma is a benign odontogenic tumor and is locally aggressive. It originates from the remnants of dental epithelium. It accounts for 1% of all tumours of the jaw. It is the second most common odontogenic tumour after odontoid. Approximately 80% of ameloblastomas occur in the mandible and 20% occurs in the maxilla.[1] Ameloblastic carcinoma (AC) is rare in mandible and in maxilla it is very unusual. Fewer than 60 cases of AC have been reported in the literature.[2] According to recent World Health Organization (WHO) classification, the primary AC demonstrates the morphological features of ameloblastoma with atypia, regardless of the presence or absence of metastasis.[3] We reported a rare case of AC in a 26-year male patient. He was treated with surgery followed by adjuvant radiotherapy.

CASE REPORT

A 26-year young male initially presented with swelling over left side maxillary area for eight months, which was gradually increased in size. Clinical examination revealed a hard, nontender, non-compressible swelling over left side maxillary region, measuring 3x4cm. Systemic examination revealed no abnormality. FNAC from the lesion was consistent with cystic inflammatory lesion. X-ray of paranasal sinuses showed opacity in the left maxillary area (Fig No.1). He was treated with partial maxillectomy. Histopathology examination

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revealed ameloblastic epithelium with peripheral palisading and ameloblastic epithelium in stroma (Fig No.2). There was increased cellularity, pleomorphism, mitosis with focal palisading and nests of tumour tissue invaded into desmoplastic stroma confirming diagnosis of AC (Fig No.3,4). The patient was received post-operative adjuvant external beam radiotherapy of 60Gy in 30 fractions, 200cGy per fraction to the tumour bed and neck region by Co60 teletherapy machine last in August-2012. Now, the patient was on regular follow-up science last two years with the disease free.

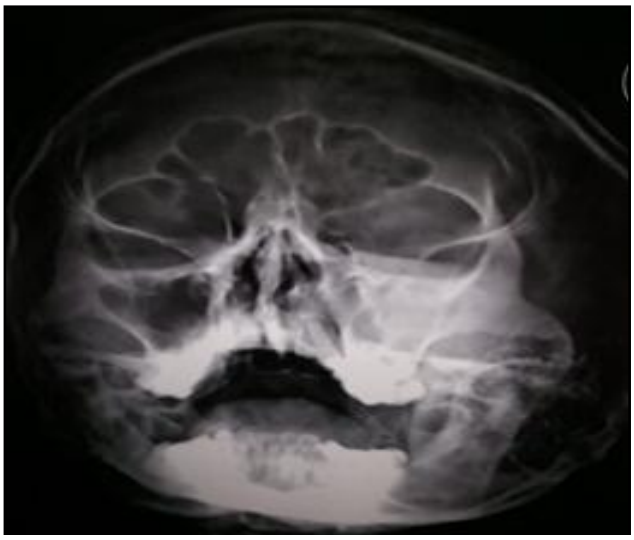

Fig No.1: X-Ray of Paranasal Sinuses Showing Opacity an in The Left Maxilla

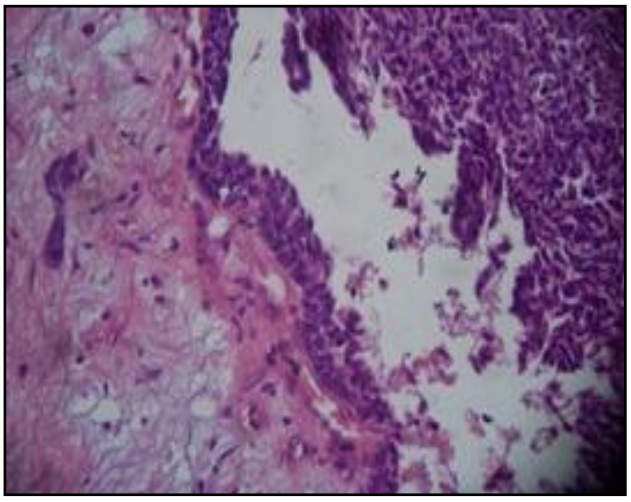

Fig No.2: Presence of Ameloblastic Epithelium with Peripheral Palisading and Nests of Ameloblastic Epithelium in Stroma (H And E, 400X).

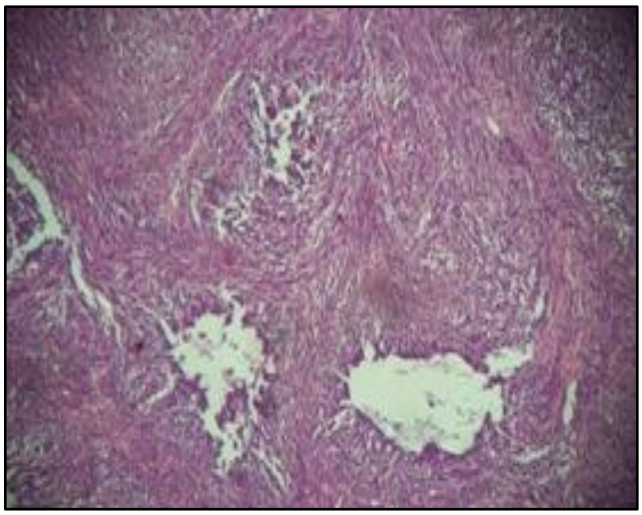

Fig No.3: Presence of Nests of Tumour Tissue Invasion into Desmoplastic Stroma in Ameloblastic Carcinoma (H And E, 40X).

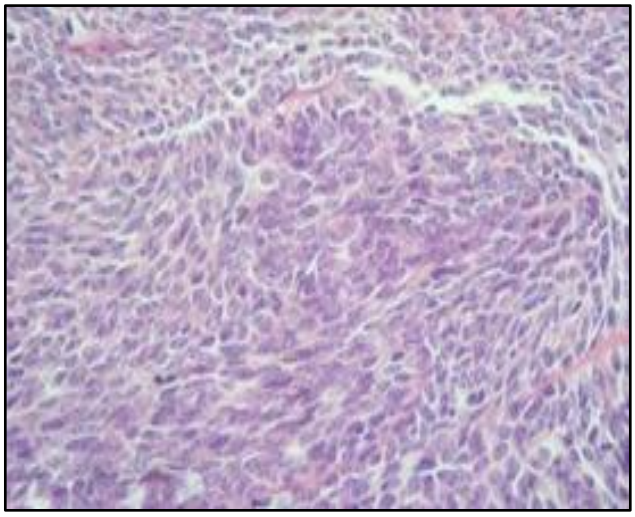

Fig No.4: Presence of Increased Cellularity, Pleomorphism and Mitosis in Ameloblastic Carcinoma with Focal Palisading (H And E, 400X).

DISCUSSION

In 1972, WHO published a classification of odontogenic carcinomas, which recognised malignant ameloblastoma, primary intra osseous carcinoma, and other cancers arising from odontogenic epithelium.[4] However, AC was not mentioned in this classification. Elzay modified the classification, but many pathologists did not agree with the proposed subgrouping.[5] The term AC was first introduced by Shafer et al in 1983.6 A malignant epithelial odontogenic tumour,

histologically retains the features of ameloblastic differentiation, and exhibits cytological features of malignancy in the primary or recurrent tumor.[6] In 2005, WHO revised the classification of odontogenic carcinoma and described the primary AC with histological features of ameloblastoma with cytological atypia with or without metastasis.[3,6] Despite these classifications, the diagnosis, histologic features, treatment, and prognosis remains challenging to clinicians. There is a lack of pathological features to distinguish ameloblastoma with no malignant/metastatic potential from malignant histology that may metastasise. Often, pathologists report ameloblastoma as a primary intra osseous carcinoma. In our case, FNAC revealed suggested of cystic inflammatory lesion, but postoperative histopathological examination revealed AC. So, biopsy is mandatory in such case before treatment.

The treatment of and prognosis for AC is unclear in the literature due to the rarity of this tumour and the lack of well-documented cases. Surgical excision, with or without adjuvant radiotherapy is required for local control. Surgery is the optimal treatment, although the best approach remains controversial.[7] For this patient, external beam radiotherapy was planned according to postoperative histopathology report of AC. As ACs are rare, there is no consensus for their treatment. Despite the lack of adequate clinical data, surgery followed by radiotherapy seems to be the treatment of choice.[8] Preoperative radiotherapy has been suggested to decrease the tumour size and may be used to treat some rapidly growing tumours before radical surgery.[9] At the University of Florida, three patients with AC were treated with surgery and postoperative radiation, to a total mean dose of 68Gy. All patients had local control after 24 months.[10] There are limited data for the efficacy of radiation. Recently published studies highlight that radiation can induce regression but not cure.[11] The role of chemotherapy is not yet proven.[12]

Currently, most clinicians treat AC in the same manner as other oral cavity cancers, with surgery and postoperative radiation. Documented reports with meaningful follow-up are rare. Meticulous follow-up is essential to ruled out recurrence and metastasis to the lungs and regional lymph nodes.[13] The present case was treated with partial maxillectomy followed by postoperative adjuvant radiotherapy. The patient was disease free since last two years of treatment.

CONCLUSION

AC of maxilla is a rare odontogenic tumour. Its histological characterization is difficult, and many differential diagnoses must be excluded. Surgical excision, with or without adjuvant radiotherapy are required for local control. These tumours must be removed with wide margins to avoid local recurrence, which frequently occurs after minimal surgical treatment. The prognosis is dominated by the risk of local recurrence as well as the distant metastasis. Systemic assessment of the chest through periodic imaging is recommended in the management and follow-up setting.

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