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An unusual case of Ameloblastic carcinoma in an Elderly Patient: A Case Report

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Abstract: *Ameloblastic carcinoma is an odontogenic rare malignant tumor. It is a locally invasive aggressive neoplasm that can spread to regional lymph nodes or remote sites such as lung and bones. AC's clinical course is usually aggressive, with comprehensive local bone destruction. Swelling, pain, trismus, important bone resorption with mobility of the tooth, dysphonia and intraoral fistula are the most prevalent clinical characteristics. Histopathology is a essential tool for ameloblastic carcinoma diagnosis and is also needed to distinguish it from ameloblastoma. Its biologic behavior recommends necessity of early diagnosis and prompt treatment. However, due to lack of case reports with long-term follow-up, there is no established treatment protocol for these cases. Here, we are presenting a unique case of ameloblastic carcinoma of left mandible in a 68-year old male patient treated successfully with resection along with the 2.4mm titanium reconstruction plate.*
Keywords: Ameloblastic carcinoma, odontogenic carcinoma, odontogenictumor

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I. Introduction

Ameloblastic carcinoma (AC) is a rare odontogenictumor that accounts for 1.5 % –2.0 % [1] of all odontogenictumors. Ameloblastoma malignant versions are extremely rare and may result from de novo or conversion of a long-standing mainly benign lesion that has undergone several surgical excisions[2]. Two-thirds of these cancers come from the mandible, while one-third come from the maxilla[3]. The most prevalent symptom is a painful swelling that is progressing rapidly[4]. It can also occur as a clinically benign cystic lesion or as a big tissue mass with ulceration, important is extensive bone resorption, and mobility of the tooth[5].

More than 3,600 instances of ameloblastomaswere described in the literature[6], but less than 60 cases of ameloblastic carcinoma were recorded, including two-thirds in the mandible[7].

There is still controversy about both the aetiology of this rare carcinoma and whether this form of carcinoma originates from an ameloblastoma or constitutes a distinct entity[7]. This rare odontogenictumor has histological tumor of both ameloblastoma and carcinoma. There are differences of view concerning the therapy of ACs; however, wide-ranging surgical excision with or without radiotherapy is the most frequently used method of treatment[8].

II. Case Report

A 68-year old male patient reported to the department of oral and maxillofacial surgery AMU, Aligarh, with chief complaint of small swelling over left side of lower face and mild pain & tingling sensation in lip since last 2 months.

Patient was asymptomatic since last 6 months earlier he noticed a painless swelling over the left side of the lower jaw. He consulted a dentist, where some medications were advised. After few days he realized tingling and numbness in lower lip . He reported to the department 3 months after that . Patient belongs to middle class socioeconomically and has no habit of smoking, and tobacco chewing.(Fig.1)

On examination a mild swelling was present over the left body region of mandible extending from 1st premolar to 2nd molar region. Patient was edentulous with slight depression in the area of alveolar ridge in relation to lesion. Mouth opening was 42mm with no deviation during movement of mandible . overlying skin was normal in colour and texture. The swelling was mildly tender on touch, hard in consistency, smooth with no regional lymphadenopathy.(Fig.2)

Radiological examination showed U–shaped bony defect, unilocular, radiolucent shadow with scalloped borders extending from mandibular left 1st premolar to 2nd molar . CT scan showed, lesion involving left body of mandible measuring 6× 4.5 × 6.0 cm in size, cortical breach in both side. (Figs. 3,4,5)

Incisional biopsy of the lesion was done and histological examination on Hematoxylin&Eosin stain revealed nest of ameloblastic epithelium with surrounding myxoidstroma epithelium showing multi layering at places.FIbrocollagenous tissue containing islands of epithelium arranged in basaloid, plexiform and occasional follicular pattern. Few epithelial islands are showing clear cell changes. The tumor cells exhibited malignant features like nuclear hyperchromatism, altered nuclear cytoplasmic ratio and many mitotic figures.(Fig.6) On the basis of above features, a diagnosis of ameloblastic carcinoma with clear cell changes was given.

On the basis of report of biopsy segmental mandibulectomy (first premolar to 2nd molar) was done taking safe margins of 1 cm under general anesthesia. The submandibular incision was given and the tumor was removed with normal margins along with the 2.4mm titanium reconstruction plate.(Fig.7) Hemostasis achieved and closure done in layers. Chemotherapy and radiation was not advised. No metatasis reported during the 1.5 year follow up period.(Figs.8,9)

III. Discussion

Ameloblastoma is an odontogenic jaw tumor that results from embryonic dental remains, potentially from an odontogenic cyst's epithelial lining; dental lamina or enamel organ; or displaced epithelial remains[5] . For many years, the malignant type of ameloblastoma was contentious. Despite their benign histology, the word ' malignant ameloblastoma' means that lesions metastasize. The word ameloblastic carcinoma (AC) is reserved for ameloblastoma with a malignant morphological appearance, irrespective of metastasis[4].

ACs are rare odontogenic neoplasms and may be caused by de novo or pre-existing odontogenic lesions[8,9]. They usually include approximately 80% in the mandible and less frequently about 20% in the maxilla[4]. It is present in a wide variety of age groups. There's no obvious predilection for sex. Mostly it is asymptomatic & swelling is the most common sign, though other signs include related pain, rapid growth, paraesthesia of surrounding soft tissue, trismus and dysphonia[10].Metastatic disease screening should be carried out, particularly in recurrent instances of typical ameloblastoma, malignant ameloblastoma, and ameloblastic carcinoma[11].

The AC's radiographic appearance is consistent with that of an ameloblastoma except for the existence of certain focal radiopacities, reflecting apparently dystrophic calcifications. In standard ameloblastomas, these histological and radiological characteristics are usually not seen.

Clinically, more aggressive than typical ameloblastomas. Cortical plate perforation, extension to surrounding soft tissue, multiple recurrent lesions and metastases, generally connected with cervical lymph nodes, may be correlated with ameloblastic carcinomas[10].

Squamous cell carcinoma, especially the basaloid variant, was the primary differential diagnosis for this tumor. In this situation, the characteristics that differentiated AC from squamous cell carcinoma included tumor cell jigsaw puzzle-type nesting, stellate reticulum presence, and unique nest cystic degeneration. In the differential diagnosis, the diagnosis of craniopharyngioma can also be regarded, mainly because of its well-known similarities to odontogenic neoplasia and partly because of its place in the cranial base. However, because the results were characteristic of AC[4], these opportunities are excluded.

The therapy of choice is surgical resection. The safest surgical method for ensuring disease-free survival is block removal with a standard bone margin of 1–2 cm. This technique has led in lower than 15% local recurrence rates[12].There is dispute about radiotherapy in ameloblastoma and it is regarded to be a radioresistant tumor[13]. There is no well-documented proof regarding these tumors ' real radioresponsive. The authors doubt their efficacy[14]. However, Atkinson et al.[15] evaluated ten patients with ameloblastomas treated with mega-voltage irradiation retrospectively and found that ameloblastoma is not an inherently radioresistanttumor and that correctly implemented mega-voltage irradiation plays a helpful role in leadership. He also concluded that main radiation should be regarded whenever, due to local invasion or inappropriate due to medical variables, a full-surgical excision was technically hard. Recommended dosages for therapy range from 3,000 cGy to 5,000 cGy. Most ameloblastic carcinomas are intraosseous; therefore, radiation therapy efficacy must be critically considered[7].Chemotherapy does not appear to be indicated as main therapy. Such therapy results for non-metastatic disease were poor[16]. Horváth et al. recorded in an 8-year-old girl a case of mandibular AC with pleura-pulmonary and bone marrow metastasis. Five cycles of vincristine, endoxane, adriamycin, carboplatin and etoposide chemotherapy were prescribed. It led in tumor shrinking but no important impact on the pulmonary site and after 8 months the patient died. Chemotherapy's function is therefore still unpredictable[17].

There was no evidence of regional or distant metastasis in the situation described by us, but in the same tumor there was histological proof of typical ameloblastic fields and anaplastic cell foci. In addition, cellular pleomorphism and nuclear hyperchromatism occurred in the same tumor with occasional mitoses. Slootweg and

Muller[8] and Daramola et.al[18] defined a case of ameloblastoma that later showed cytological proof of malignancy in the main lesion after numerous surgeries indicating that repeated surgical trauma could be liable for malignant conversion.Multiple local recurrences, repeated operations, and radiation therapy or chemotherapy often by numerous recurrences, or the repeated surgical procedures needed to treat these recurrences may cause tumor cells to be implanted into blood vessels or lymphatic channels.

After definitive treatment, ACs may recur locally in 0.5–11 years[19]. Usually, distant metastasis is deadly and may appear postoperatively as soon as 4 months or as late as 12 years[19]. The lung is the most prevalent location for distant metastasis, followed by bone, liver, and brain[19,20]. In the lack of a local or regional recurrence, distant metastasis may occur[20].

IV. Conclusion

Ameloblastic carcinoma is an unusual unit of odontogenictumors in main or distant metastasis that displays malignant histological characteristics. This is in comparison to malignant ameloblastoma where there are benign histological characteristics of both main and distant metastasis. In patients with toothache or mobile teeth in combination with constant jaw swelling, pain, and fast development, it is essential to consider it as a differential diagnosis. Prognosis is good if early diagnosis is performed with prompt surgery, but in neglected cases where metastasis is distant, prognosis is poor.

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Conflicts of interest
There are no conflicts of interest.

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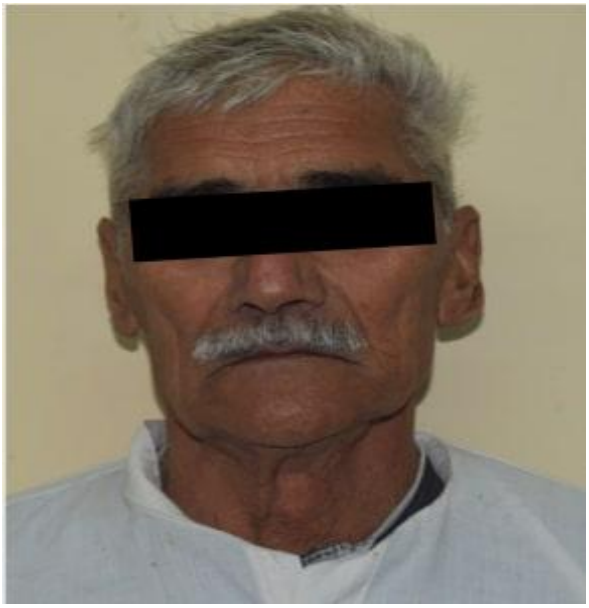

Fig.1: Preoperative photograph of the patient showing slight swelling at left side.

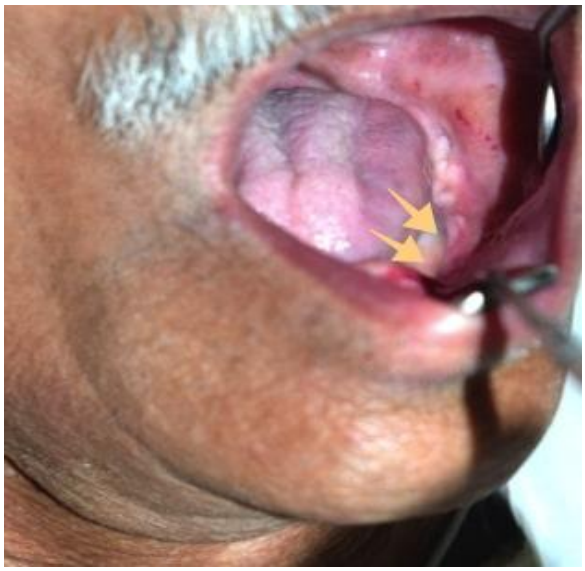

Fig.2: Intraoral findings at the first visit. Both jaws were edentulous, with obliterated vestibule and U- shaped deformity in left side of body of mandible.

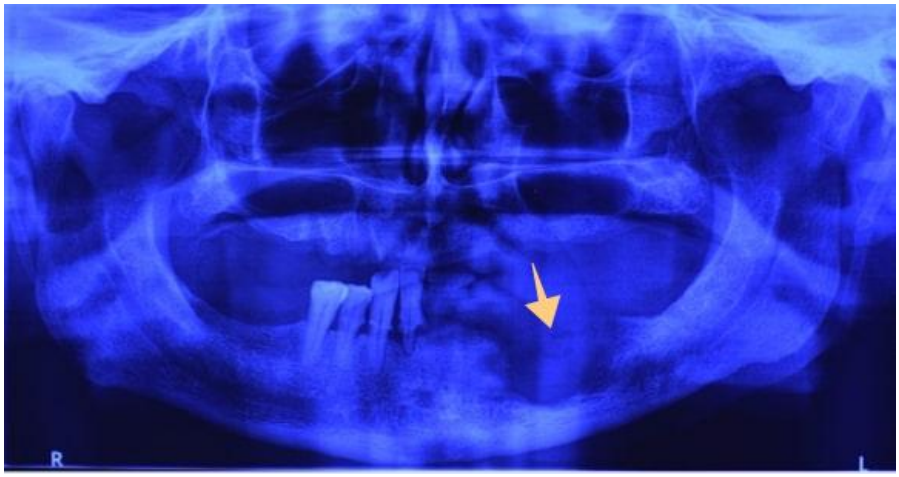

Fig.3: OPG showing radiolucency at left side

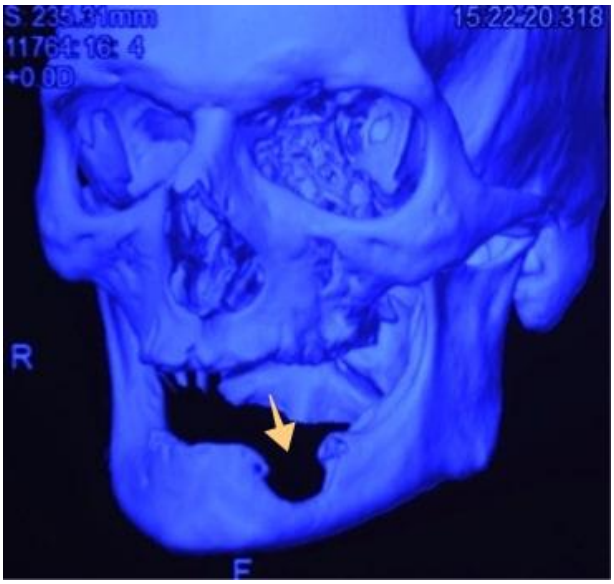

Fig.4: 3D view Computed tomogram showing u-shaped bony deformity in left side of mandible.

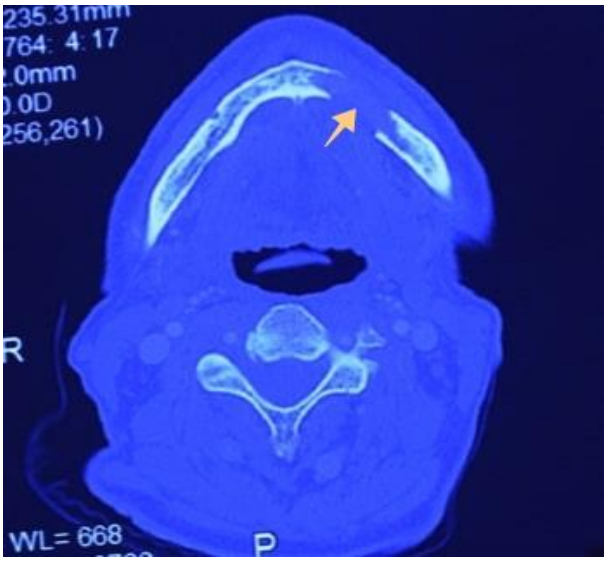

Fig.5: Axial view of Computed tomogram showing bone lacking continuity in both lingual and buccal cortex.

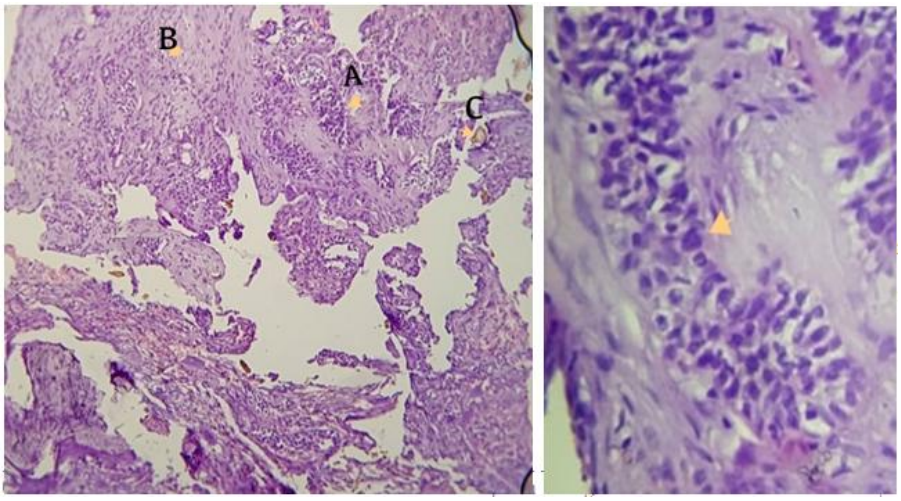

Fig.6: Photomicrograph showing (A) odontogenic epithelial island with ameloblast-like cells (b)Epithelial islands showing clear cell changes.(C) necrotic bony tissue seen in stroma. Photomicrograph showing nuclear hyperchromatism and pleomorphism.

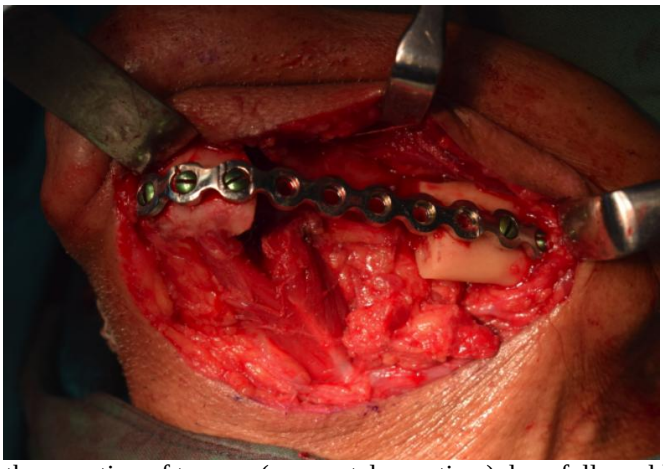

Fig.7: Operated site after the resection of tumour (segmental resection) done followed by placement of 2.4 mm titanium reconstruction plate

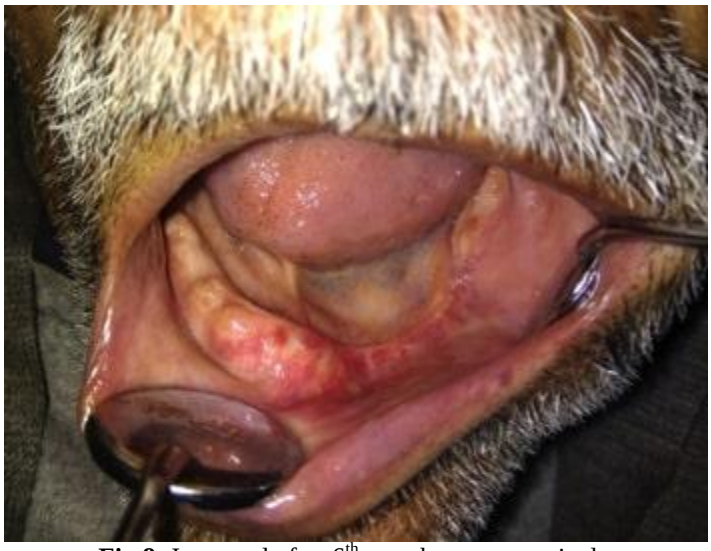

Fig.8: Intra oral after 6th month post operatively

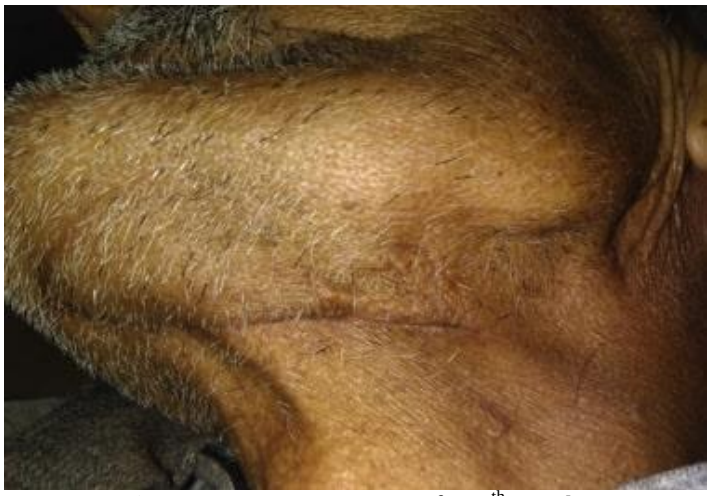

Fig.9: Post operative image after 6th month.

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