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Ameloblastic Carcinoma-Secondary Type,A Rare Case Report and Distinction from Malignant Ameloblastoma

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CASE REPORT

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ABSTRACT

Ameloblastic Carcinoma (AC) primarily is an aggressive odontogenic tumour, which histologically consist of features of both ameloblastoma and carcinoma. This requires more aggressive surgical approach then its benign counterpart. Malignant Ameloblastoma (MA) microscopically appears benign but shows metastasis. The authors reported an extremely rare secondary type of Mandibular Ameloblastic Carcinoma of a 50 year old Indian male with a complaint of swelling, trismus, significant bone resorption with tooth mobility. This article discussed about the clinical features, radiographic features, management, and histological characteristics of AC and its distinction from MA.

INTRODUCTION

Ameloblastic Carcinoma (AC) is a rare malignant odontogenic tumour that has features of ameloblastoma intermingled with features of carcinoma, regardless of whether it has metastasized [1] i.e., ameloblastoma in which there is histologically malignant transformation with less differentiated metastatic growth [2]. Malignant Ameloblastoma (MA) is the lesions that metastasizes and are well differentiated benign lesions AC in both primary and metastatic sites [3]. In contrast to ameloblastoma, presents more aggressive clinical features, such as rapid growth, perforation of cortex, painful swelling and trismus [4]. AC mostly occurs in posterior mandible region with only 1/3rd cases in maxilla region, and mostly occurs in around 30 years of age [5-10]. Radiographic findings include a poorly defined radiolucency, sometimes with focal radiopacities due to necrosis with dystrophic calcifications [7,8]. According to latest WHO 2005 update, (AC) are classified in to Primary type i.e., histologic features of ameloblastoma with cytologic atypia and Secondary type, i.e., malignant transformation of pre-existing benign ameloblastoma [11]. Most cases of (AC) are denovo and less than 1% of ameloblastomas undergo malignant transformation. Hence secondary type is less frequent than primary [12-15]. In this article, a case of (AC) secondary type is presented and its clinical examination, radiographic examination, histopathology and management are discussed. Meanwhile the published articles about the (AC) are reviewed and its distinction from MA is pointed.

CASE PRESENTATION

A 50-year-old Indian male patient referred to our department with the complaint of rapidly growing painless swelling of the right side of lower jaw crossing midline with difficulty in chewing, multiple mobile teeth over affected region. He had the past history of a similar lesion over the right lower jaw, which was treated by Hemimandibulectomy with Disarticulation of right TMJ 12 years back. The histopathology then was suggestive of ameloblastoma. On extraoral examination, a firm diffuse non-tender swelling over right side of f(Ac)e extended superiorly from the right infraorbital region to the lower border of mandible inferiorly. Medially, it extended up to one-third of both upper and lower lips with an elevated angle of mouth and laterally 1 inch anterior to the angle

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of mandible with more medio-lateral expansion measuring about 10 cm × 8 cm × 6 cm **(Figure 1A)**. Neither paresthesia was associated with the swelling nor was any regional lymph node palpable. Intraoral examination revealed a painless firm proliferative growth crossing midline up to the right anterior border of ramus. The swelling was negative on aspiration **(Figure 1B)**.

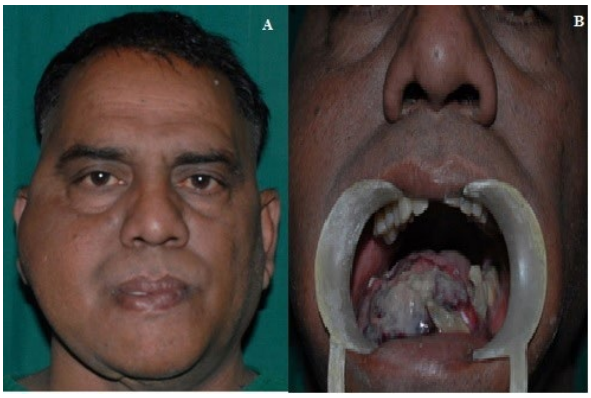

Figure 1. A: Recurrence after year following surgery, **B:** Ulcero proliferative growth in the mandibular anterior region.

Radiographic Examination

3 Dimensional computed tomography showed hypodense image of approximately 5 cm × 7 cm size present over left side mandibular and submandibular region, an area of resorption over left mandibular premolar region with attached 2.4 mm titanium reconstruction plate **(Figure 2)**.

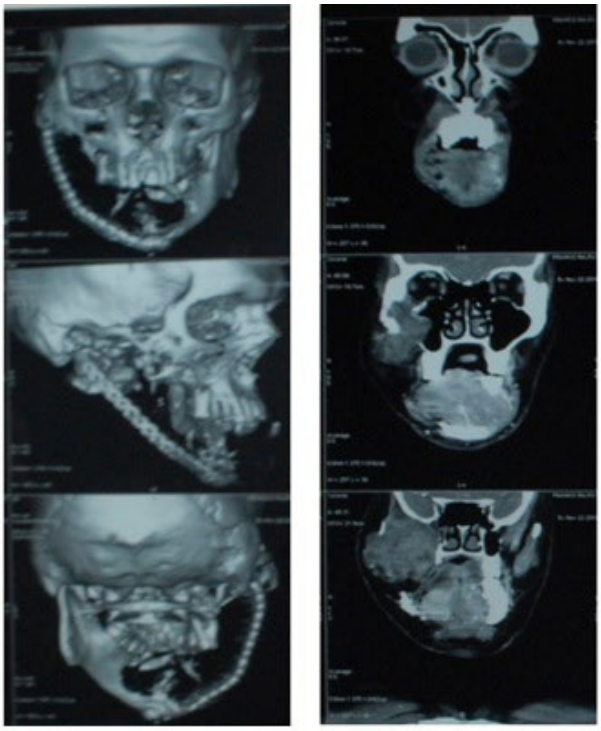

Figure 2. Computed tomographic scan shows bony destruction of left mandible crossing midline.

Incisional Biopsy

The patient was then planned for an incisional biopsy under local anesthesia. Histopathological examination with H and E stain was conclusive of diagnosis of AC.

Surgery

He was planned for resection of tumor under general anesthesia. The submandibular incision was given and the tumor appeared firm in consistency, surrounding the reconstruction plate. The tumor was removed along with the reconstruction plate and reconstruction was done with Pectoralis major myocutaneous flap **(Figure 3)**. Hemostasis (Ac)hieved and closure done in layers. The postoperative recovery was uneventful.

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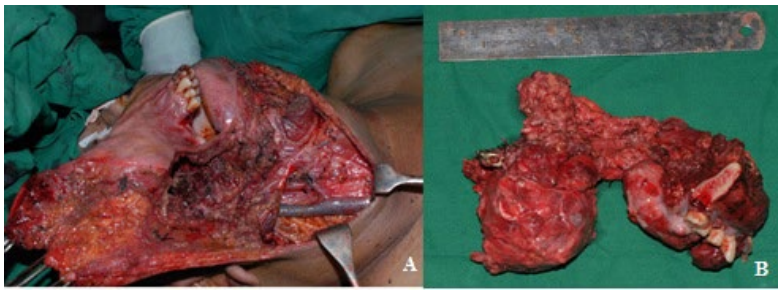

Figure 3. A: Operated site after the resection of tumour, **B:** Resected surgical specimen.

Histopathology

Microscopic examination showed ameloblastlc islands with palisading of peripheral cells showing reverse polarity. Within the islands marked features of malignancy were seen that is increased mitosis, hyperchromatism, and pleomorphism. Some of the islands lost its arrangement of peripheral palisading columnar cells and exhibited irregularly arranged squamous cells with mitotic figures, vesicular nuclei and prominent nucleoli **(Figure 4)**.

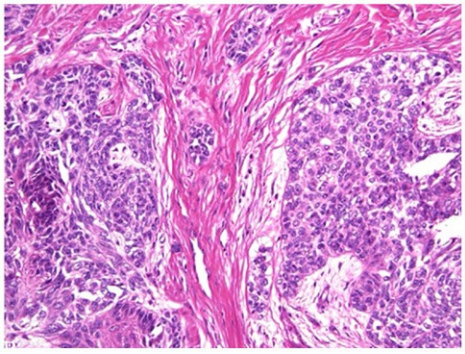

Figure 4. Histological examination with hematoxylin-eosin, original magnification X 20.

Follow Up

Then we referred the patient to department of radiotherapy. They have given 25 fractions of 1.8 Gy e(Ac)h for a period of 5 weeks (45 Gy). Then they referred the patient to Department of Pulmonary Medicine and Gastroenterology to evaluate distant metastasis, reports was negative. The patient is on regular follow-up since 1 year and doing well.

DISCUSSION

In 1983, Shafers introduced the term (AC), which is a rare malignant neoplasm that requires correct histologic diagnosis and aggressive surgical intervention. The tumor cells resemble the cells seen in ameloblastoma, but they show cytologic atypia. Mean time span from symptoms to treatment is shorter for (AC) (11 months) as compared with benign ameloblastoma (27 months) as its progression is faster [16,17]. Most (Ac), primary type, arise de novo and histologically defined as an apparent malignant neoplasm that exhibits cytological malignancy, including pleomorphism, hyperchromatic nuclei, and high mitotic rates. The rare variety of (Ac), secondary type, is thought to occur from the recurrent or pre-existing ameloblastoma, and it may have the histologic features of a coexisting ameloblastoma [12,15,18]. The clinical features, biologic behavior, and treatment of (AC), secondary and primary types, are almost identical. Because of its rarity, no full proof therapeutic guidelines exist for (AC). Some clinicians recommend wide resection. However a small number of reports have described radiation therapy and chemotherapy as treatment modality with Ion beam therapy as a new measure.

After reviewing all article on AC present in Pubmed and Cochraine online digital library till date we got that AC mostly occurs in sixth to seventh decade of life. They are seen twice as often in male patients as in female patients. The most commonly involved area is the posterior portion of the mandible [19-26]. Involvement of the maxilla by (AC) seems to be less frequent [25]. The most common sign described has been swelling, although others include associated pain, rapid growth, trismus and dysphonia [26]. The radiographic appearance of the lesion was consistent with that of an ameloblastoma except for the presence of some focal radiopacities, apparently reflecting dystrophic calcifications [26].

MA is a very rare tumor, defined in 1992 by the World Health Organization (WHO) as “a neoplasm in which the pattern of an ameloblastoma and cytologic features of malignancy are shown by the primary jaw lesion and/or by any metastatic growth”. The term MA was especially controversial because of the former WHO classification, in which the diagnosis was based on the presence of metastasis. Histologically, MA does not show uniform proliferation, but it has cellular pleomorphism. Spindling of the cells is

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recognized in some solid proliferating areas [26].

The two forms of malignancy described in association with ameloblastomas are the MA and (AC). For a diagnosis of MA, the cardinal feature is metastatic spread, because the histological appearance of the primary and metastatic lesion(s) is indistinguishable from the typical benign ameloblastoma. The most common sites of metastases are reported to be the chest (lungs, pleura, hilar nodes) in 75% of cases and the cervical lymph nodes or spine in 15% [24]. The mode of spread of MA is debated as being either by inhalation at the time of surgery, haematogenous or lymphatic spread. The management of MA [19,20]. Isolated and discrete lung metastases have been treated by open thor(Ac)otomy and wedge resection 6V15 and chemotherapy has occasionally been used successfully [21-23].

CONCLUSION

In the present case the lesion is confined clinically and radiographically, within the mandible with evidence of extension of a carcinoma into gingiva, alveolar ridge, floor of mouth, but no evidence of metastasis to distant site. Microscopically, there is carcinoma as well as morphologic features that resembled ameloblastoma, arose in a preexisting ameloblastoma. These clinical, radiographic, pathologic findings support the diagnosis of an (AC) secondary type. Only a limited number of (AC)s secondary type cases have been reported in the English-language literature till 2017, so this present report adds one more case to the scanty number of publication.

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