



## A Rare Case of Ameloblastic Carcinoma of Mandible and Its Reconstruction with Iliac Crest Graft

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### Abstract

*Malignant odontogenic tumors are rare and represent approximately 1% of all oral malignancies. Ameloblastic carcinoma is a rare malignant lesion with characteristic histological features and behaviour that dictates a more aggressive surgical approach than that of a simple ameloblastoma. It occurs primarily in the mandible in a wide range of age groups. It may arise de-novo or in pre-existing ameloblastoma or odontogenic cyst. The clinical course of ameloblastic carcinoma is typically aggressive, with extensive local destruction. Direct extension of the tumor, lymph node involvement and metastasis to various sites (frequently the lung) have been reported. Wide local excision is the treatment of choice. This article describes a rare case of a ameloblastic carcinoma of the mandible encountered in a 28 year-old female patient and includes an insight into reconstructive procedures in such situation.*

**Keywords-** Ameloblastic carcinoma, Metastasis.

### Introduction

Odontogenic malignancies are rare lesions arising from dental embryogenic residues. Odontogenic carcinomas have been designated by a variety of terms, including malignant ameloblastoma, ameloblastic carcinoma (AC), metastatic ameloblastoma, or primary intra-alveolar epidermoid carcinoma. The clinical course and histopathology of these tumours are similar to each other, so the differential diagnosis is not easy to make. Malignant ameloblastoma represent tumours that metastasize while both primary and metastatic

lesions retain their benign histological appearance. The term ameloblastic carcinoma was introduced by Elzay<sup>1</sup>. In the last update of the WHO classification, published in 2005<sup>2</sup>. it is defined as a rare odontogenic malignancy that combines the histological features of ameloblastoma with cytological atypia, even in the absence of metastasis. Two thirds of these tumors arise from the mandible while one third originates in the maxilla<sup>3</sup>. The most common symptom is a rapidly progressing painful swelling. It may also present as a cystic lesion with benign clinical features or

as a large tissue mass with ulceration, significant bone resorption and tooth mobility<sup>4</sup>. Both etiology of this rare carcinoma and the question whether this type of carcinoma originates from an ameloblastomas or represents a separate entity are still controversial<sup>5</sup>. There are difference of the opinion regarding treatment of ameloblastic carcinoma (AC), however, wide surgical excision with or without radiotherapy is the most commonly used treatment modality<sup>6</sup>.

### Case Report

A patient, age 28 years, female, came to our centre with chief complaint of swelling, pain and parasthesia on the right side of lower jaw (Fig.1). Curettage was done one year ago at some other centre but since past few weeks patient again complaints of pain and swelling in the same region. Swelling was present on the right side of face. Overlying skin was normal, no visible pulsation, no discharge were seen, swelling was hard, tender, no local rise in temperature, mouth opening and jaw movements were satisfactory. On intraoral examination, there was rounded bony swelling of the posterior alveolar bone. No ulceration was found, mucosa was normal, 3<sup>rd</sup> molar was missing. Orthopantomogram revealed Multilocular radiolucent region starting from right 1<sup>st</sup> incisor to third molar of right side of mandible (Fig.2). Incisional biopsy of the lesion was done and histological features were suggestive of Ameloblastic Carcinoma. The patient was taken up for surgical resection and primary stage mandibular reconstruction with iliac crest graft under general anesthesia. A segmental resection was done followed by reconstruction with iliac crest graft and reconstruction plate and screws (Fig.3). Chemotherapy and radiation was not advised. No metastasis reported during the 2 year follow up period.

### Discussion

Ameloblastic carcinoma is a rare neoplasm that represents a challenge in its diagnosis, treatment, and prognosis. Information regarding its clinical

features is scanty. Ameloblastic carcinoma has been reported to arise either de novo, ex odontogenic cyst, or ex ameloblastoma. Majority originates de novo, and the remaining are malignant transformation of an ameloblastoma. According to WHO 2005 classification<sup>2</sup>, carcinomas derived from ameloblastomas are designated as: 1) Metastazing malignant ameloblastoma; 2) Ameloblastic carcinoma (a) Primary, (b) Secondary (dedifferentiated) intraosseous, (c) Secondary (dedifferentiated) extraosseous. Malignant ameloblastoma histologically shows the features of benign ameloblastoma, but it metastasizes to the other sites such as lung. Ameloblastic carcinoma will have the features of cytological atypia along with the histological features of benign ameloblastoma in the primary tumour, irrespective of the presence or absence of metastasis<sup>7</sup>. In the case presented by us, there was no evidence of regional or distant metastasis but there was histological evidence of typical ameloblastic areas and foci with cellular pleomorphism and nuclear hyperchromatism with occasional mitoses in the same tumour. Surgical resection is the treatment of choice. En bloc removal with 1–2 cm of normal bone margin is the safest surgical modality to ensure disease-free survival. This method has resulted in local recurrence rates of less than 15%<sup>8</sup>. Cervical lymph node dissection should be considered when there is obvious lymphadenopathy. There is controversy regarding radiotherapy of Ameloblastoma, and it is considered radioresistant tumour<sup>9</sup>. No extensive studies have been presented which demonstrate the role of chemotherapy in treatment. Treatment in this case consisted of segmental resection with 1.5 cm margins and defect was reconstructed with nonrevascularised iliac crest bone graft. Several methods exist for reconstruction of segmental mandibular defects: nonvascularised bone grafts (NVBGs), titanium reconstructive splints, or free flap transfers that allow the use of vascularised bone. Among the latter, iliac crest, scapula and fibular flaps are most widely used for mandible reconstruction. As suggested by Foster et al<sup>10</sup> non

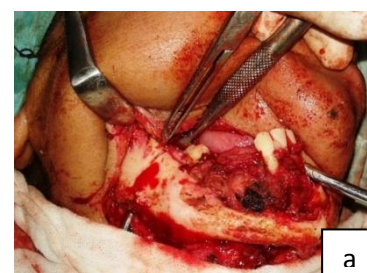
revascularised bone grafts are utilized in those cases with bone defect inferior to 5–6 cm usually without interruption of bone continuity, good blood supply and soft tissue bearing. Revascularised bone grafts are indicated in all those cases that show a large bone defect (more than 6 cm), with interruption of the bone continuity or with maintenance the cortical aspects, but with soft tissue compromise. They also reported 17% failure for 6 cm defect and 75 % for 12 cm defect for NVBG. Although VBFs (free vascularised fibular flap has the least resorption and failure rate) are at present a first choice treatment for mandibular reconstruction, Disadvantages include the inability to reproduce adequate height of alveolar bone, inability to reconstruct large soft tissue defects, inability to use in the presence of peripheral vascular disease, and decreased vascularisation when a large number of osteotomies are required. Corticocancellous graft from iliac crest used for the reconstruction of mandible provided good contour with excellent bulk and height necessary for implant rehabilitation. The limitations of NVBG lies in the fact that it is avascular which makes it susceptible to infection and increases the chances of failure with increasing length of defect. NVBGs such as iliac crest graft still have a place in mandibular reconstruction. We had an acceptable functional and esthetic outcome in this particular case with satisfactory graft uptake, uneventful healing, and no immediate or delayed complications. Long-term follow-up is ongoing. Ameloblastic carcinoma is a rare entity of odontogenic tumors that exhibits malignant histologic features in the primary or metastasis. ACs can recur locally 0.5–11 years after definitive therapy. One should be alert for the possibility of local recurrences and distant metastases especially to the lung, bone, or brain. A regular assessment of the chest by periodic imaging is recommended.



**Fig;1** showing extra oral swelling On right side



**Fig; 2** Multilocular radiolucent region starting on right side of mandible.



**Fig.3** (a).Intraoperative appearance of lesion,  
(b) Resected specimen  
(c) Reconstruction with iliac crest graft and reconstruction plate

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