

## Case Report

### Ameloblastic carcinoma: Report of a Case

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#### Abstract

Ameloblastic carcinoma is a rare epithelial odontogenic tumour of the jaws which exhibits cytological features of ameloblastoma and carcinoma. A case of ameloblastic carcinoma in a 52 year old male is reported. Clinical / histological characteristics of this tumour and current knowledge on the classification of odontogenic malignancies are discussed. The importance of including ameloblastic carcinoma in the differential diagnosis of persistent jaw swellings associated with toothache or mobility of teeth or failure of healing of extraction sockets is emphasized.

#### Introduction

Ameloblastoma is a benign but locally aggressive neoplasm, which clinically presents as a slowly growing painless swelling of the jaws. Although it is reported to constitute about 1-3% of all jaw tumours and cysts<sup>1</sup> it is the most frequently encountered odontogenic tumor in our environment.<sup>2,3,4,5</sup> Reports from the literature suggest that it is more common in Blacks than in Whites<sup>6,7</sup>. Its malignant variants are exceptionally rare and may arise de-novo or from transformation of a long-standing primarily benign lesion which has undergone several surgical treatment<sup>8</sup>. Although the terms malignant ameloblastoma and ameloblastic carcinoma have been used interchangeably for these variants in the past, it is now generally agreed that malignant ameloblastoma tends to metastasizes in spite of the benign histological characteristics of the primary jaw lesion while ameloblastic carcinoma is an ameloblastoma

Clinical examination revealed a moderate ill-defined, bony hard swelling of the left cheek. The swelling was firm, non tender, not cystic, not warm and not attached to overlying skin. There was no associated anaesthesia. The left submandibular nodes were firm, slightly enlarged but not fix. Intraoral examination also revealed a missing lower left first molar with an exophytic soft tissue growth from the residual socket. Intraoral mandibular swelling extended from the lower left first bicuspid to the retromolar area with bucco-lingual expansion of the jaw. Associated lower

showing histological evidence of malignancy in the primary recurrent or metastatic lesion<sup>9</sup>. Both variants must also have evidence of rapid clinical growth. In view of the rarity of these tumours, report of cases to encourage documentation of their clinical characteristics and response to treatment has been advocated<sup>10</sup>. We present a case of ameloblastic carcinoma in a 52 year- old man which exhibited both histological characteristics of ameloblastoma and carcinoma. Case Report

A 52 year old business man presented in our Dental Clinic in February 2002 with a 3 month history of left mandibular swelling. He had earlier on attended a clinic in Saudi Arabia for the same problem and he claimed a biopsy was carried out which indicated an Ameloblastic carcinoma. Swelling was preceded by toothache following which a tooth was removed in September 2001. This resulted in failure of healing of the extraction socket and a soft tissue growth over the socket 2 months post-extraction.

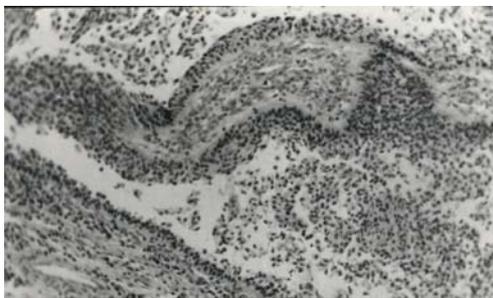
left premolars and molars were mobile. Extra-oral plain radiographs revealed a unilocular radiolucency of the left mandibular body with ill-defined margins. Chest Xray and further clinical examination failed to reveal any metastatic disease.

A repeat incisional biopsy was done and the initial report indicated acanthomatous ameloblastoma but on further a report of ameloblastic carcinoma was reached. Patient was operated under G.A. on 30<sup>th</sup> April 2002. A composite block resection of the left mandible from canine region to the ascending ramus

just above the angle was carried out with the sparing of the superior half of the ascending ramus. Mandibulo-maxillary fixation was used to stabilize the jaws for 4 weeks.

The resected specimen measured 6 x 5x4 cm and weighed 130g. There was subcutaneous tissue to a depth of 1.5cm and the left mandible. A grey-tan tumour was present within the mandible with a variegated cut surface. Microscopy of the resected specimen showed irregular masses and interdigitating cords of epithelial cells with palisading around the periphery of the epithelial islands. The tissue in the center of the cellular islands composed of stellate reticulum. Other areas show island of keratinizing well-differentiated squamous cell carcinoma infiltrating the adjacent bone. The draining submandibular lymph nodes exhibited reactive

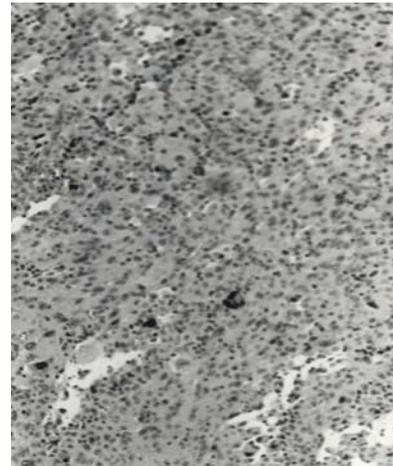
**Fig 1 Stromal invasion by sheets of palisading cells.**



hyperplasia. Post operative course of antibiotics included Ceftriazone (Rocephin™) and metronidazole for five days.

The post-operative period was uneventful and patient was discharged home after 4 weeks. Patient is being followed up and as at the last review (2 years post-op), was tumour free. Secondary reconstruction via iliac bone graft was planned but the patient was not psychologically prepared to undergo a second surgical operation.

**Fig2 Well differentiated SCC.**



SCC=squamous cell cancer

## DISCUSSION

Malignant epithelial odontogenic tumours which include malignant ameloblastoma, ameloblastic carcinoma, primary intraosseous squamous cell carcinoma, clear cell odontogenic tumour, and malignant epithelial ghost cell tumour are very rare. They are said to arise from the rest cells of Malassez, or Serres, reduced enamel epithelium or linings of epithelial odontogenic cysts.<sup>(1)</sup> Until recently a lot of controversies existed on the classification of this group of tumours. Pindborg et al.<sup>(12)</sup>, classified malignant epithelial odontogenic neoplasms and other tumours related to the odontogenic epithelium (odontogenic carcinomas) into three main groups which are: A. Malignant ameloblastoma B. Primary intraosseous carcinoma C. Other carcinomas arising from odontogenic epithelium including those arising from odontogenic cysts. This classification grouped carcinomas arising from odontogenic cysts with those from odontogenic tumours and did not consider histological delineations.<sup>(13,14)</sup> Elzay<sup>(13)</sup> in a review of primary intraosseous carcinomas suggested a modification which would allow the recognition of ameloblastic carcinoma as an entity separate from carcinomas arising from cystic linings. The modification he proposed is as follows: Odontogenic Carcinomas: Type 1 : Arising from odontogenic cysts. Type 2 : Arising from ameloblastoma a) Well-differentiated (malignant ameloblastoma) b) Poorly

differentiated (ameloblastic carcinoma) Type 3 : Arising de-novo I Non-keratinizing type ii Keratinizing type. Slootweg and Muller<sup>(14)</sup> further modified this classification as follows: Type 1: Primary intraosseous carcinoma arising from odontogenic cyst Type 2: A - Malignant ameloblastoma B -Ameloblastic carcinoma arising de-novo, ex- ameloblastoma and odontogenic cyst. Type 3: Primary intraosseous carcinoma arising de-novo A. Non-keratinizing B. Keratinizing. According to the latest revision of World Health Organization's (WHO) classification<sup>(15)</sup>, ameloblastic carcinoma is in the group C of odontogenic carcinomas. That is, malignant variants of other odontogenic tumours thus taking care of previous criticisms. Nagai and colleagues<sup>(16)</sup> in reviewing odontogenic carcinomas reported in the English literature prior to 1991 reclassified 46 cases as ameloblastic carcinoma using the criteria advocated by Slootweg and Muller<sup>(14)</sup>. Few additional cases have been reported since then<sup>9,10, 16,17,18,19,20</sup> The consensus now is to use the term ameloblastic carcinoma for those tumours with histological evidence of malignancy in the primary, recurrent, or metastatic tumour regardless of whether there is metastasis or not while malignant ameloblastoma is reserved for metastasizing ameloblastomas which exhibit benign histological features both in the primary and metastatic lesion<sup>20</sup>.

In the present case, there was no evidence of regional or distant metastasis but there was histological evidence of typical ameloblastic cells and anaplastic cells in the same tumour (Fig 1). In addition, there was cellular pleomorphism and nuclear hyperchromatism with occasional mitoses in the same tumour (Fig 2). Slootweg and Muller<sup>14</sup> reported a typical case of an ameloblastic carcinoma arising from a pre-existing ameloblastoma after many repeated surgeries and radio-therapeutic treatment were carried out for recurrent episodes. Daramola et al.<sup>(21)</sup> also described a case of recurrent maxillary ameloblastoma which later exhibited cytological evidence of malignancy in the primary lesion after multiple surgeries suggesting that repeated trauma caused by surgery could be responsible for the malignant transformation.

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Although we could not ascertain unequivocally whether ameloblastic carcinoma in this patient developed de-novo or from a pre-existing ameloblastoma, we believe the former might be the most likely due to the absence of any history of previously operated tumour from the site and the short duration of the lesion. Wide resection is the acceptable treatment option for jaw malignancies. Immediate jaw reconstruction is not routinely done in our centre due mainly to the fact that frozen-section is not available to check the status of the surgical margins intraoperatively. A delayed bone graft was planned but up till the time of this report the patient was yet to be convinced since he has been able to cope adequately and his present facial appearance is acceptable

In conclusion, although odontogenic carcinomas are rare, it is important to rule out these malignancies in patients who present with toothache or mobile teeth in association with persistent jaw swelling or failed healing of extraction sockets through prompt radiological and histopathologic investigations. This will encourage early and prompt treatment which improves prognosis

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