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Ameloblastic Carcinoma – A case report highlighting its variations in histology

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Abstract:
The term "Ameloblastic carcinoma (AC) is defined as an ameloblastoma in which there is histological evidence of malignancy in the primary tumour or the recurrent tumour regardless of metastasis"
The frequency of malignant behaviour in ameloblastoma is difficult to determine but probably occurs in far less than 1% of all ameloblastomas.
This article discusses a case report which was documented as an ameloblastic carcinoma with histological variations that were observed under special stains and immunohistochemical stains. This also proves that an ameloblastoma shows a spectrum of histological and biological behaviour ranging from benignity at one end to frank malignancy at the other end.
Keywords: Ameloblastoma, pathology, carcinoma, pathology, case report.

Introduction:

Ameloblastoma is an odontogenic tumor arising from the dental embryonic remnants possibly from the epithelial lining of an odontogenic cyst, dental lamina, enamel organ, stratified squamous epithelium of oral cavity or displaced epithelial remnants.[1,2]

Its malignant transformation has been reported with rarity in the literatures. The malignancy arising from ameloblastoma leads to either a malignant ameloblastoma or ameloblastic carcinoma [3].The term malignant ameloblastoma should be used for a tumour that shows the histopathological

features of ameloblastoma both in the primary tumour and in the metastatic deposits. The term AC should be reserved for an ameloblastoma that has cytological features of malignancy in the primary tumour, in a recurrence or in any metastatic deposit. According to Cawson[4], the term AC is an ameloblastoma which shows cytological features of malignancy but is otherwise recognizable as an ameloblastoma.

A malignant tumor can be classified as odontogenic if it demonstrates epithelium that histologically resembles that seen in developing tooth or a recognizable odontogenic tumor such as ameloblastoma.[5-7]

There are various classification systems from the literature. The recent updated World Health Organization classification of odontogenic carcinoma is

1. Metastasizing, malignant ameloblastoma
2. Ameloblastic carcinoma
   a. Primary
   b. Secondary (dedifferentiated), intraosseous
   c. Secondary (dedifferentiated), extra osseous

In this article, we present a case of an ameloblastoma showing dysplasia in the primary lesion as an ameloblastic carcinoma. Further it is also proposed to discuss some of the histological variations that were observed in the primary lesion and in the recurrence under immuno histochemical stains and special stains.

Case History:

A male patient of 55 years had reported to our outpatient department with a chief complaint of an ulceroproliferative growth in the left upper maxillary region of three months duration. (Fig. 1)

Patients had the habit of smoking for the past 15 years.

The growth was initially small, gradually increased in size and was painful. Patient had undergone extraction of 27, 28 due to mobility. On general examination the patient was malnourished. On extra oral examination a mild swelling was noticed in the middle third of the left side of the face. Intraoral examination showed poor oral hygiene. Lesional examination revealed a growth of 4x3 cm on the alveolar ridge of upper left posterior region. The lesion extended anteriorly from the distal aspect of 26 and posteriorly upto maxillary tuberosity. On palpation, the mass was firm in consistency and tender. Hence a provisional diagnosis of squamous cell carcinoma was given.

Orthopantomograph of the patient showed soft tissue shadow in the left maxillary region, poor resolution of the maxillary tuberosity with breach in the maxillary sinus. Biopsy was taken from the lesion and was subjected for histopathological examination. Microscopically the incised tissue exhibited marked pleomorphism, hyperchromatism and other features confirming the malignant nature of the epithelial origin. However some of the areas exhibited follicular configuration resembling that of an enamel organ which is unusual in a typical squamous cell carcinoma. The malignant nature of the epithelial lesion was confirmed. On this basis, radical resection was carried out and the postoperative specimen was examined in detail for further confirmation.

From the post-operative specimen multiple sections were taken and the histopathology was studied in detail. The overall histological picture was showing multiple islands individually resembling enamel organ type of tissue with palisading of peripheral tall columnar cells. The multiple enamel organ type of tissue was predominant in deeper sections (Fig. 2).

Higher magnification of the individual islands showed peripheral tall columnar cells of enamel organ type which clearly exhibited change in polarity of nucleus (Fig. 3).

However the continuity of palisading arrangement was not appreciable in some areas. The central areas of islands showed large prominent cells exhibiting malignant features in the form of hyperchromatism, increased mitotic figures like tripolar nuclei and pleomorphism (Fig. 4).

The histological picture was more consistent with follicular ameloblastoma, however it exhibited marked malignant changes, and hence histopathological diagnosis of ameloblastic carcinoma was given.

The Patient reported two months after surgery with recurrence (Fig. 5).

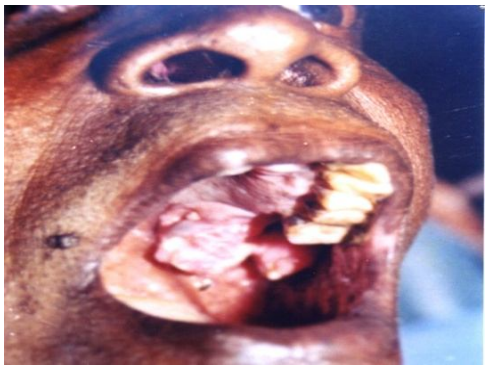

Fig 1- Ulcero proliferative growth in the left maxillary posterior region.

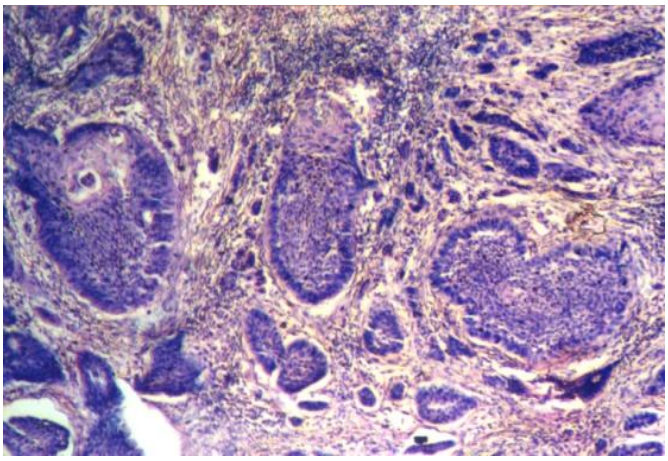

Fig 2- Multiple epithelial islands with palisading tall columnar cells (H&E 10X)

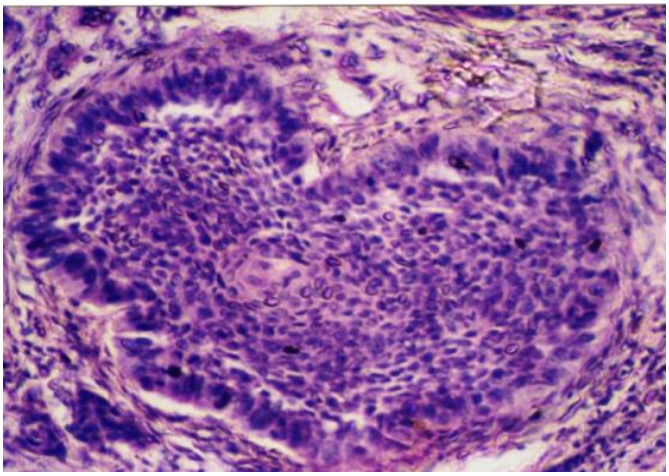

Fig 3-Epithelial Island with peripheral tall columnar cells exhibiting change in polarity of nucleus (H & E 40 X)

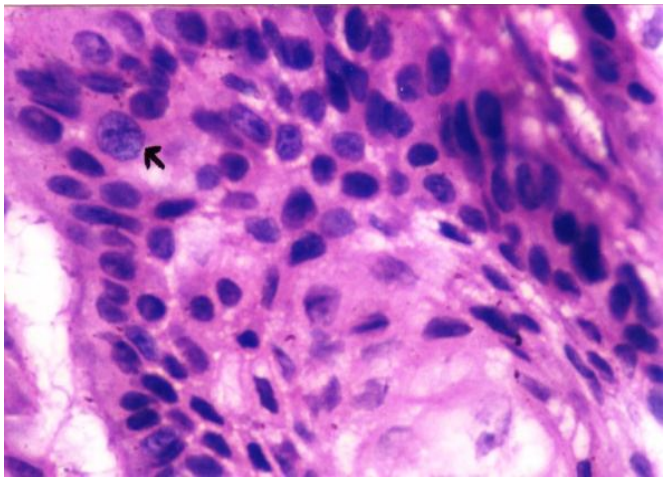

Fig4-Epithelial Island exhibiting malignant features of pleomorphism, hyperchromatism, (tripolar nuclei) and squamous transformation (H & E 40 X)

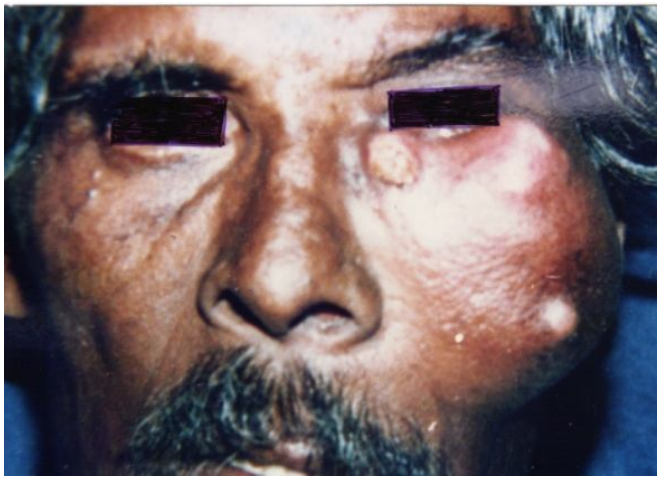

Fig 5- Recurrence after 2 months following surgery.

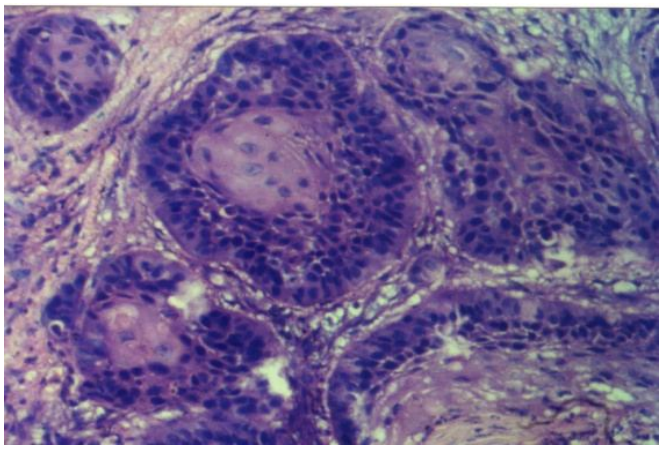

Fig 6 - Recurrent lesion with more prominent squamification of central cells of ameloblastic islands (H& E 10X)

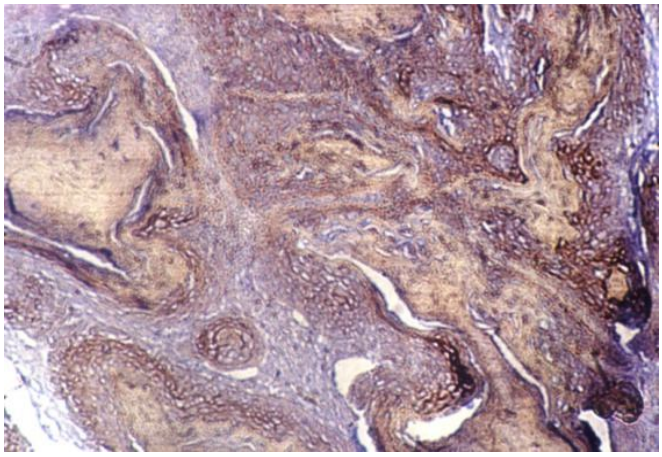

Fig 7 - Cytokeratin immuno reactivity of AEI, AE3 in areas of squamous metaplasia and weak staining in peripheral cells. (40X)

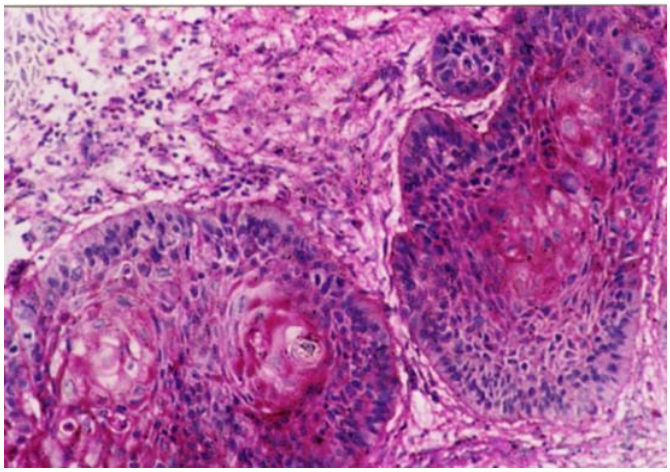

Fig 8 - PAS stain strongly reacting in central areas of islands and with discontinuity of basement membrane at areas of severe dysplasia. (PAS, 10X)

The tissue taken from the recurrent lesional area for histological examination was consistent with the postoperative diagnosis of ameloblastic carcinomas, but with more prominent features of squamous metaplasia of central cells of ameloblastic tissue (Fig. 6).

Few of the sections exhibited areas of malignant epithelial cells showing mitotic figures, vesicular nuclei and prominent nucleoli similar to the microscopic appearance of squamous cell carcinoma.

Part of the specimen was subjected for immunohistochemical examination using cytokeratin antibodies to evaluate the behaviour of the central cells. Immunohistochemical staining exhibited strong cytokeratin immune reactivity for AEl, AE3 in the areas of squamous metaplasia and weak staining in the peripheral cells (Fig. 7).

PAS staining was also carried out to see the continuity of the basement membrane in the individual islands. Generally PAS was strongly reacting in central areas of the islands while the peripheral area exhibited discontinuity of the surrounding basement membrane particularly in the areas which exhibited severe dysplasia (Fig. 8).

Discussion

The term ameloblastic carcinoma is used to designate those ameloblastomas that show histopathological features of malignancy in the primary or secondary lesion regardless of metastasis.[8]

Approximately 2/3 of AC involves the mandible. Only 19 cases have been reported to have occurred in the maxilla.[9].This case was considered unique since it exhibited different histopathological features at the various stages of progression. Initially the present case indisputably exhibited features of ameloblastoma in predominant areas in the form of islands resembling enamel organ type with characteristic palisading of peripheral cells showing reverse polarity. But within the ameloblastlc islands marked features of malignancy were noticed (viz) increased mitosis, hyperchromatism, and pleomorphism. In the recurrent lesion many 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 similar to the microscopic appearance of squamous cell carcinoma suggesting a change in the histological pattern. It is however controversial whether ameloblastic carcinomas are transforming biologically and histologically from a classical state to a frank squamous cell carcinoma. Lionel Gold et al[9] states that similar histological changes of loss of peripheral palisading cells in the islands have been noted.

As stated by Stephen et al[5] it is quite probable that, the ameloblastoma shows a spectrum of histological and biological behaviour ranging

from benignity at one end to frank malignancy at the other end. Periodic Acid Schiff staining reaction in our case also proves that there is loss of continuity of the basement membrane particularly in aggressively behaving areas.[10-12] Disruption of basement membrane may be related to more aggressive nature of ameloblastomas which only supports the views of Stephen that in later stages microscopic appearance of some of the cases of ameloblastic carcinoma can end up as squamous cell carcinoma. Bruce and Jackson,[13] also summarized the features of ameloblastic carcinoma as a neoplasm showing histological evidence of malignant transformation of the ameloblastoma like epithelium in the primary tumour, while metastases tend to resemble a less well differentiated SCC.

Our case also exhibits decreased cytokeratin positivity in the areas of marked malignancy whereas normal ameloblastic cells and well differentiated squamous cells showed strong to moderate positivity which concurs with the findings of Stephen, Hiroyuki.[14-16] .It is reasonable to understand that this case illustrates the malignant spectrum of ameloblastoma. Cases of ameloblastoma should thus be studied carefully, correlating their histological pattern with biological behaviour to detect subtle changes in histology.

Conclusion:

The case reported here underlines the fact that even if an initial diagnosis of squamous cell carcinoma of the jaws is made, any presence of islands resembling enamel organ type should be probed further for evidence of features to support the possibility of a transformation from an already existing ameloblastic carcinoma to a squamous cell carcinoma as has been proved in our case.

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