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A Multi-Faceted Ameloblastic Carcinoma: A Rare Case Presentation

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ABSTRACT: **Background:** Ameloblastoma is a benign but locally aggressive neoplasm that rarely presents a frank malignant behaviour with the development of metastases. The frequency of malignant behaviour in ameloblastomas is difficult to determine but the transformation occurs supposedly in less than 1% cases. The term ameloblastic carcinoma is reserved for an ameloblastoma that has cytologic features of malignancy in the primary tumor, in a recurrance or in any metastatic deposit.

Case presentation: A 66 year old female patient reported with a chief complaint of a painless extra-oral swelling on the right cheek region. On palpation a firm, exophytic, sessile lesion with fixation to the underlying tissues was seen. Crackling egg sign was also elicited. Radiography revealed a multi-locular lesion involving the right side of the body and the anterior border of the ramus of the mandible. The clinical diagnosis was given as mural ameloblastoma. Excisional biopsy was performed, followed by histopathological examination of the specimen. Carcinomatous features like cellular pleomorphism, nuclear hyprchromatia, anisocytosis and anisonucleosis, dyskeratosis were seen in sheets of proliferating malignant epithelial cells in few areas. Whereas certain areas revealed desmoplastic, granular, acnthomatous, follicular and plexiform patterns.

Conclusion: Ameloblastic carcinoma is quite a well reported entity in medical literature. The uniqueness about this case is multiple patterns juxtaposing with carcinomatous areas which again resembles squamous cell carcinoma. A knowledge of this will increase the diagnostic aptitude of pathologists.

KEYWORDS: Ameloblastoma, Ameloblastic carcinoma

I. INTRODUCTION

Traditionally, ameloblastoma has been regarded has a benign tumor, that can be locally aggressive, but occassionally metastasize, resulting in death. Thus the term 'metastatic ameloblastoma' has been used for lesions showing ameloblastoma-like features in distant tissues due to such metastasis. Typically, the primary ameloblastic lesion occurs in the molar ramus area of the mandible in young adults, preferably from 2nd to 3rd decade of life[1]. The average age of presentation has been found to be 30 years with about 33% of patients aged at or below 20 years of age. Metastatsizing ameloblastomas show metastatic deposits are seen in the lungs, cervical lymph nodes or extragnathic bones[2]. Histopathology reveals no significant differences with respect to features, except for the aforementioned sites, confirming silent metastases.[1][2][5-7]

Ameloblastic carcinoma is a malignant epthelial proliferation, which may either be associated with an existing ameloblastoma, or may arise as de novo. The nomenclature for the former is carcinoma-ex-ameloblastoma, whereas that of the latter is de-novo ameloblastic carcinoma[23]. It represents a greater degree of cellular atypia, as compared to malignant ameloblastomas, alongwith increased mitotic activity. Ameloblastic carcinomas are locally agrressive neoplasms, that may spread to regional lymph nodes, or distant sites, such as lungs and extragnathic bones.

Carcinoma ex ameloblastomas arise directly from pre-existing ameloblastomas, when there is de-differentiation causing clones with less differentiation and high proliferative capacity[22]. The high proliferative property overgrows the benign ameloblastoma and becomes the dominant component[4-6]. A follicular or plexiform ameloblastoma blends through a narrow hypercellular zone with sheets of mitotic bodies with hyperchromatic nuclei and basaloid shape, larger squamoid cells with pale vesiculated nuclei or elongated spindled epithelial cells. Mild to moderate pleomorphic nuclei are also seen[11].

De-novo ameloblastic carcinomas lack a conventional primary ameloblastic lesion and its categorization as ameloblastic carcinoma is less secure and is a highly subjective diagnosis[12].

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II. CASE HISTORY

A 66 year old female patient reported with a chief complaint of a painless extra-oral swelling on the right cheek region. The lesion developed approximately 5 years ago, with a small nodular swelling on the alveolar bone on the right molar ramus region. The lesion increased in size over time, with no additional symptoms. A biopsy performed about 2 years ago revealed revealed ameloblastoma diagnosis, however, no excision was performed. There was no relevant medical or familial history, and no habits were found.

III. CLINICAL FEATURES

General physical examination, along with a detailed local extra-oral and intra-oral examinations were performed. The patient was of obese built, with normal gait, and no cyanosis, jaundice, koilonychia, clubbing or oedema was elicitated. The vital signs recorded were also within normal limits.

Local examination was done both extra-oral and intra oral. The extra-oral examination revealed a gross facial asymmetry with respect to the right cheek region, and no other abnormalities were detected in light of lips and temporomandibular joint. The mouth opening was also normal, recorded at 48 mm.

Intra-oral examination was done for both hard and soft tissues. Teeth 44 to 46 were missing, consistingof the hard swelling in the residual alveolar bone, along with vestibular obliteration. The intra-oral soft tissue showed no deviations from normalcy. On palpation a firm, exophytic, sessile lesion with fixation to the underlying tissues was seen. Crackling egg sign was also elicited. No associated tenderness, paraesthesia or other symptoms were seen. No indurated borders were noted.

IV. RADIOGRAPHIC FEATURES

Radiography, done by cone-beam computed tomography (CBCT) revealed a multi-locular lesion involving the right side of the body and the anterior border of the ramus of the mandible. The clinical diagnosis was given as mural ameloblastoma. Excisional biopsy was performed, followed by histopathological examination of the specimen.

V. HISTOPATHOLOGY

Carcinomatous features like cellular pleomorphism, nuclear hyprchromatia, anisocytosis and anisonucleosis, dyskeratosis were seen in sheets of proliferating malignant epithelial cells in few areas. Whereas certain areas revealed desmoplastic, granular, acnthomatous, follicular and plexiform patterns.

VI. CASE SUMMARY AND DISCUSSION

A 66 year old female patient reported with a chief complaint of a painless extra-oral swelling on the right cheek region. On palpation a firm, exophytic, sessile lesion with fixation to the underlying tissues was seen. Crackling egg sign was also elicited. Radiography revealed a multi-locular lesion involving the right side of the body and the anterior border of the ramus of the mandible. The clinical diagnosis was given as mural ameloblastoma. Excisional biopsy was performed, followed by histopathological examination of the specimen. Carcinomatous features like cellular pleomorphism, nuclear hyprchromatia, anisocytosis and anisonucleosis, dyskeratosis were seen in sheets of proliferating malignant epithelial cells in few areas. Whereas certain areas revealed desmoplastic, granular, acnthomatous, follicular and plexiform patterns.

VII. TREATMENT AND PROGNOSIS

Conservative surgical excision is the only probable line of treatment. Radiotherapy and Chemotherapy may prove effective in reduction of tumor size before excision. Regular follow ups are advisable.

The overall prognosis as concluded by several authors is found to be poor with less than 34% 5-year survival rate, with highest survival recorded at 11 years[5][7][9-11][13-23].

VIII. CONCLUSION

Ameloblastic carcinoma is quite a well reported entity in medical literature[12-20]. The uniqueness about this case is multiple patterns juxtaposing with carcinomatous areas which again resembles squamous cell carcinoma. A knowledge of this will increase the diagnostic aptitude of pathologists.

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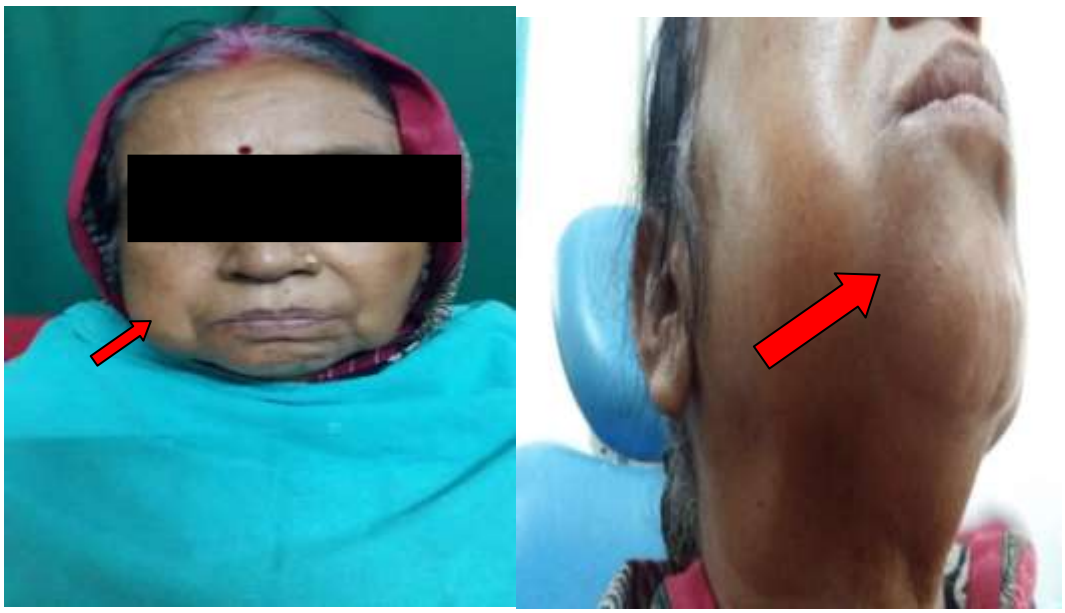
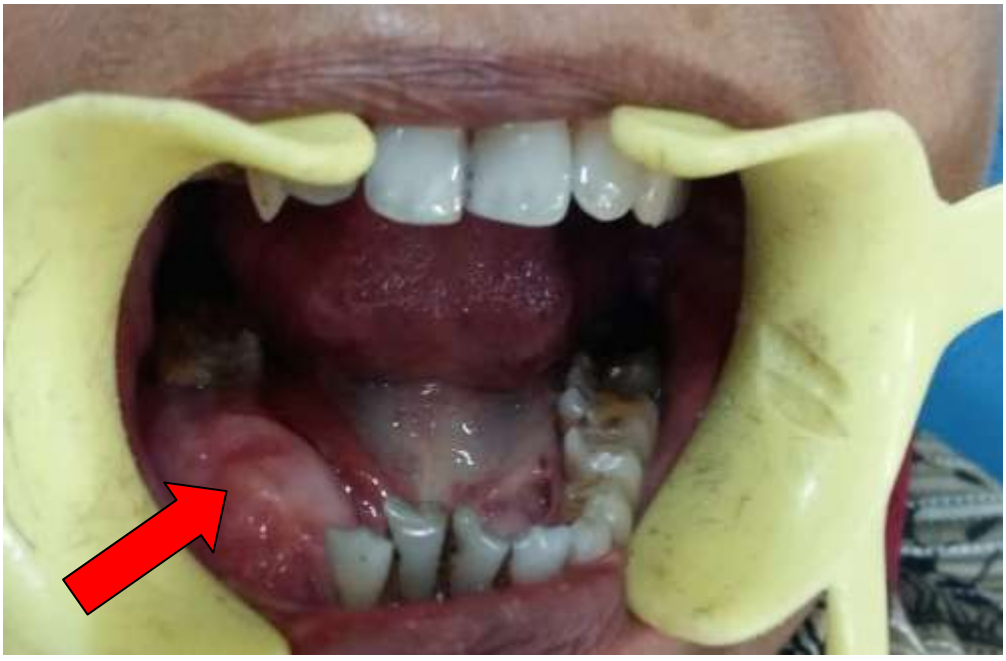


IMG-1(A) &1(B): EXTRA ORAL SWELLING wrt RIGHT CHEEK REGION



IMG-2: INTRA ORAL EXAMINATION SHOWING SWELLING wrt 43 TO 46

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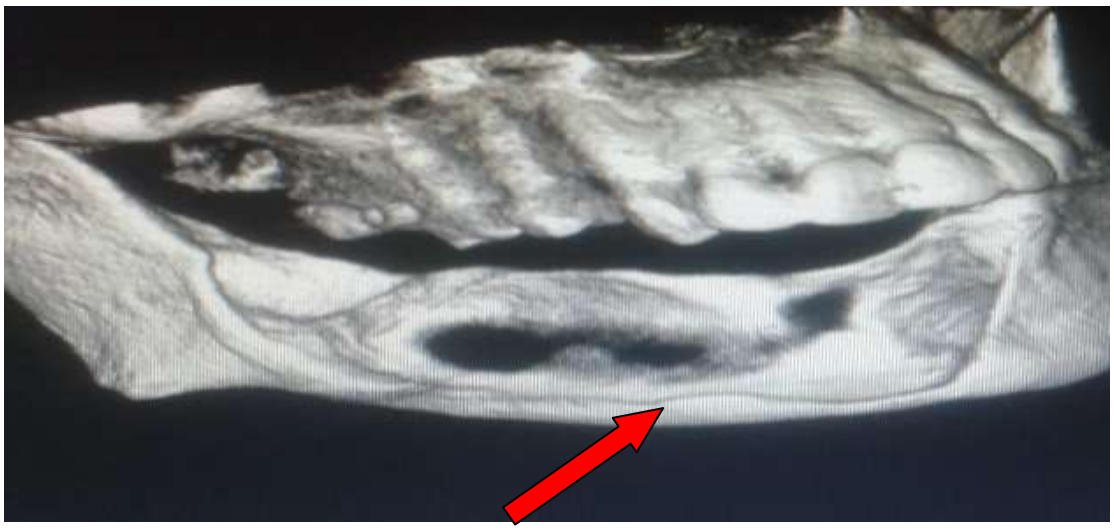

IMG-3: CONE BEAM COMPUTED TOMOGRAPHY OF AFFECTED SITE SHOWING MULTI-LOCULARITY

IMG-4: GROSSING AFTER EXCISIONAL BIOPSY ALONG WITH EXTRACTION OF MOBILE TEETH

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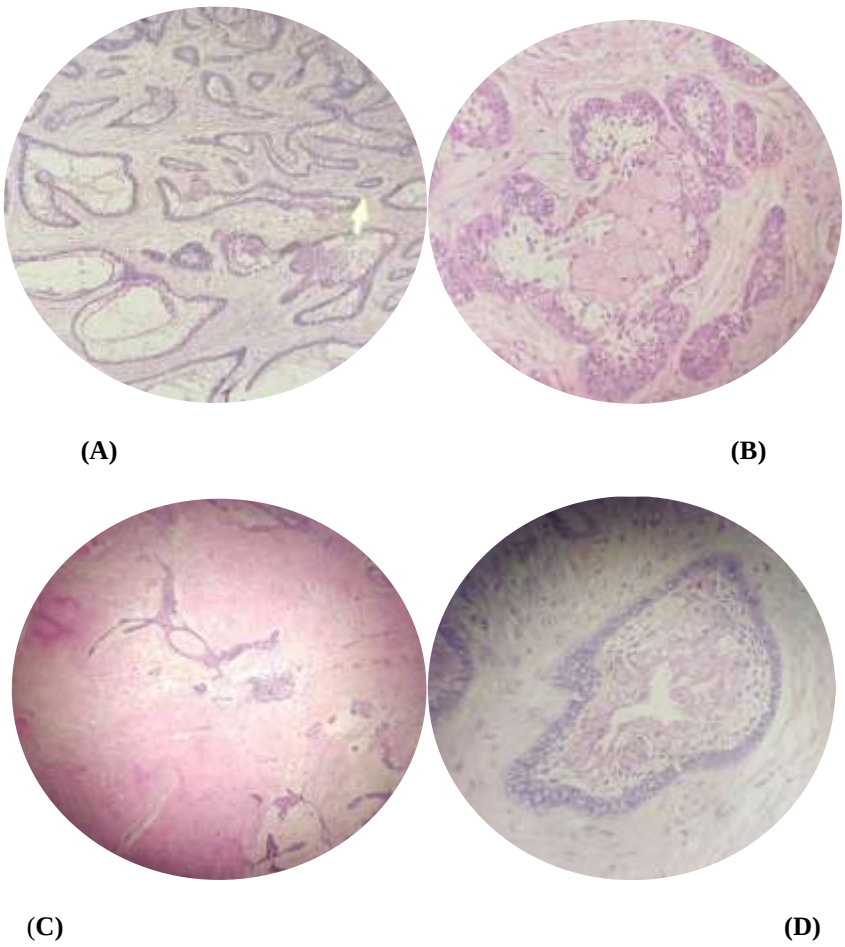


(A) **(B)**

(C) **(D)**

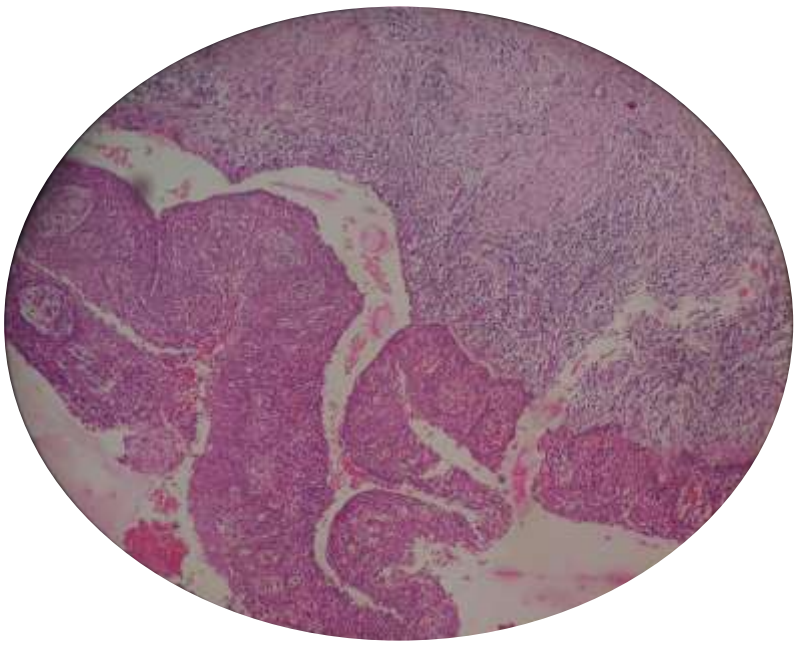
IMG-5: HISTOPATHOLOGICAL SECTIONS SHOWING ALLNORMAL VARIANTS OF AMELOBLASTOMA FEATURES

A- FOLLICULAR

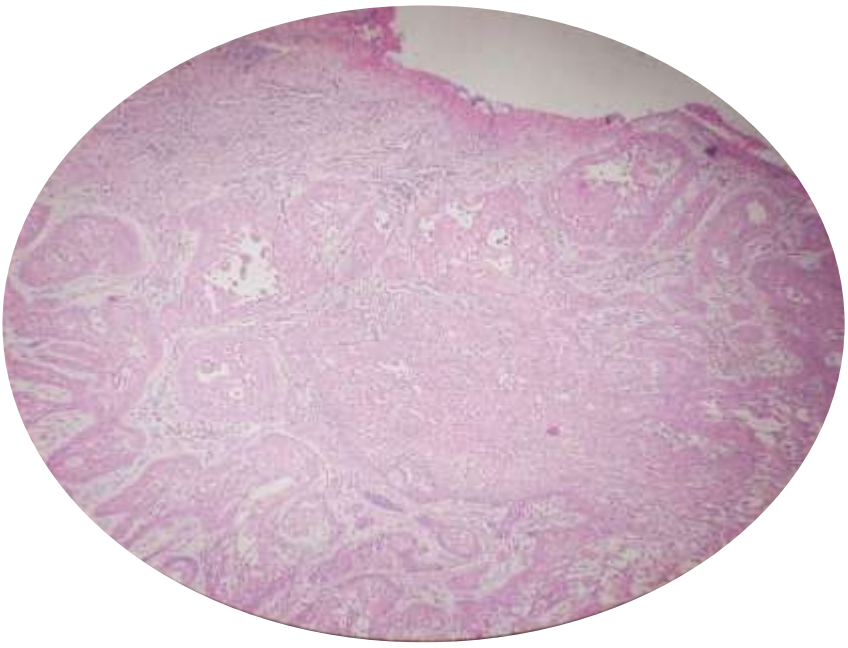
B- GRANULOMATOUS

C- DESMOPLASTIC

D- SQUAMOUS METAPLASIA



(A)



(B)

IMG-6: (A) & (B)-FRANK CARCINOMATOUS CHANGES SEEN IN SPECIMEN SECTION SHOWING DYSPLASTIC AND PROLIFERATIVE FEATURES

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