Ameloblastic Carcinoma of Mandible: A Case Report

Owais Gowhar1, Narendra Nath Singh2, Tasneem S Ain3, Saima Sultan4
1Dental Surgeon, Department of Dentistry, Government Jammu and Kashmir Health & Family Welfare Department, Jammu and Kashmir, India,
2Professor & Head, Department of Oral Pathology & Microbiology, Kothiwal Dental College & Research Centre, Moradabad, Uttar Pradesh, India,
3Consultant, Department of Public Health Dentistry, Kothiwal Dental College & Research Centre, Moradabad, Uttar Pradesh, India, 4Consultant, Department of Pedodontics & Preventive Dentistry, Kothiwal Dental College & Research Centre, Moradabad, Uttar Pradesh, India

Ameloblastic carcinoma is an uncommon malignant odontogenic tumor of jaws and is a highly aggressive tumor with rapid growth and also a high potential for distant metastasis. The most common site of occurrence is posterior mandible. Clinically, it is aggressive and has potential for local destruction. The majority of the cases arise de novo, while few cases arise from a pre-existing ameloblastoma. The radiological feature of the ameloblastic carcinoma is usually similar to that of ameloblastoma except for the presence of focal radiopacity, which apparently reflects dystrophic calcification. Herein, we report a case of ameloblastic carcinoma in the left mandible of a 60-year-old female patient.

Keywords: Ameloblastoma, Ameloblastic carcinoma, Odontogenic tumor

INTRODUCTION

Ameloblastic carcinoma is a rare epithelial odontogenic tumor of the jaws, which demonstrates all features of ameloblastoma except that it has malignant cytology.1 On the other hand, metastatic ameloblastoma, which is a very rare condition, shows minimal cytologic atypia but has undergone metastasis.2 Ameloblastic carcinoma is mostly encountered in third to fifth decade of life with male predilection. It is associated pain, mucosal ulceration, and paresthesia of affected region.3 Mostly, more than two-thirds cases occur in posterior mandible, however, in maxilla cases also have been reported.4 The majority of ameloblastic carcinomas arise de novo while as those that arise from pre-existing ameloblastomas are considered secondary or dedifferentiated lesions.5

These tumors exhibit follicular which is most common, plexiform, or trabecular pattern; infiltrative islands of tumor cells exhibit peripheral cell palisading and reverse nuclear polarization with central stellate reticulum like areas that are hypercellular; comedo necrosis, mitotic activity, and pleomorphic cells are present, and sometimes, peripheral palisading is lost; Ki-67 index is high; clear cells, ghost cells, and keratinization may be seen with focal dentinoid production.6

Primary intraosseous carcinoma and de novo ameloblastic carcinoma are likely related lesions, with the former showing less ameloblastic differentiation, in that basal cell palisading or formation of stellate reticulum-like areas is minimal or insignificant.7

CASE REPORT

A 60-year-old female came to the Department of Oral Pathology and Microbiology, Kothiwal Dental College and Research Centre, Moradabad in 2014, with a chief complaint of swelling on the left side of the face for the last 6 months. Swelling was associated with mild pain, and the patient had difficulty in mastication and mouth opening. The patient also gave a history of mobility of 2nd and 3rd molar teeth in the third quadrant which she had undergone extraction 6 months back.

Clinical examination revealed overlying skin was normal in color and texture and on palpation the swelling was mildly tender, smooth, and uniformly bony hard in consistency. A diffuse swelling over the left body ramus region of the mandible (Figure 1) which extended anteroposteriorly from the left corner of mouth to the pre-auricular region. Superiorly, the extent was up to the infraorbital region and inferiorly to the submandibular region. The margins were not well defined, and paresthesia was associated with the swelling and regional lymph nodes were not palpable. Mouth opening was 30 mm with no deviation of mandible noted. Introraomal examination revealed a diffuse swelling of the left lower buccal vestibule extending from the canine region to the retromolar area (Figure 2). Intraorally, the swelling was firm to hard in consistency and mildly tender.

Corresponding Author:
Dr. Owais Gowhar, MDS (Oral Pathology and Microbiology), J&K Health and Family Welfare Department, Directorate of Health Services, Kashmir, India. E-mail: drowais83@gmail.com
Orthopantomograph revealed radiolucency which was well-defined involving the left body, angle and ramus of the mandible (Figure 3). Chest radiograph was advised which ruled out any metastatic deposits. An incisional biopsy was done (Figure 4), and the tissue was sent for histopathologic examination which revealed anastomosing cords of odontogenic epithelium. The cords of epithelium were bounded by columnar ameloblasts like cells with nucleus reverse polarized. Epithelial cells were hyperchromatic with dysplastic features. Extensive squamous metaplasia within the stroma was evident. In some areas, microcyst formation was also appreciated within the epithelial islands. Overall histopathological features were suggestive of ameloblastic carcinoma (Figure 5).

**DISCUSSION**

Ameloblastomas account for 1% of all tumors and cysts in the jaw bones and are considered benign odontogenic tumors. These odontogenic tumors mainly occur in the mandible while maxillary ameloblastomas represent only approximately 1-20% of all cases. Histologically benign, ameloblastomas are related to 2 malignant tumors such as malignant ameloblastomas and ameloblastic carcinomas.
Malignant ameloblastoma shows the histopathologic features of an ameloblastoma both in the primary tumor and in the metastatic deposits while as the term ameloblastic carcinoma has cytologic features of malignancy in the primary tumor, in the recurrent tumor, or in any metastatic deposit.9

Ameloblastic carcinoma is aggressive and responsible for the extensive destruction of local structures. The most common clinical findings are swelling, associated pain, rapid growth, and trismus.10 Radiological findings show ill-defined radiolucency. However, focal radiopacity may be detected in radiolucent lesions.11 Microscopically, it resembles the features of ameloblastoma, except for the epithelium, which shows various cytological features of malignancy. Ameloblastic carcinomas have characters of both a benign ameloblastoma and carcinoma. A palisade arrangement of epithelial cells with nuclei away from the basement membrane, i.e., reverse polarity, is a characteristic feature of benign ameloblastomas. The epithelial cells of ameloblastic carcinomas have histopathological features of hyperchromatism, a high mitotic rate, and a high nuclear-to-cytoplasmic ratio.12

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Regarding the differential diagnosis of ameloblastic carcinomas, two types of typical ameloblastoma must also be considered. The acanthomatous ameloblastoma which demonstrates varying degrees of squamous metaplasia and keratinization of the stellate reticulum portion of the tumor islands but peripheral palisading is maintained with no cytologic features of malignancy are seen.7 Keratoameloblastoma is rare that include prominent keratinizing cysts that may cause some doubt while diagnosing the said lesion. Squamous cell carcinoma arising in the lining of an odontogenic cyst histologically resembles oral squamous cell carcinoma than ameloblastic carcinoma.7

Squamous odontogenic tumor, another differential, it epithelium lacks cytologic evidence of malignant disease and islands of squamous epithelium lack stellate reticulum like zones and peripheral palisading. In addition, microcystic changes and dystrophic calcifications may be seen in this lesion.7

CONCLUSION

Ameloblastic carcinoma is an uncommon neoplasm of jaw that combines the histological features of an ameloblastoma with features of cytological atypia. Cases of ameloblastomas should be studied thus cautiously to detect subtle changes in histology that may predict aggressive behavior and bad prognosis of these lesions.

REFERENCES