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International Journal of Orofacial Biology

Case Report

Ameloblastic Carcinoma of Anterior Mandible - A Case Report

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How to cite: Vikraman S.,Ameloblastic Carcinoma of Anterior Mandible – A Case Report. Int J Orofac.Biol.2024; 8(1):29-34.

DOI: *https://doi.org/10.56501/intjorofacbiol.v8i1.1138*

Received: 22/04/2024 *Accepted:30/04/2024* *Web Published:28/05/2024*

Abstract:

Odontogenic malignancies are rare and have spectrum of clinical presentations, making diagnosis challenging. Ameloblastic carcinoma(AMCa) is a rare odontogenic malignant lesion with characteristic clinical and histopathological features. Histologically it resembles ameloblastomatous tumor cells with cellular atypia. In this presentation, we are going to discuss a case of ameloblastic carcinoma of the anterior mandible in a 75-year-old male patient with a typical aggressive clinical course which posed a diagnostic difficulty due to variable histopathological presentation.

Keywords: ameloblastoma, odontogenic tumors, carcinoma.

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INTRODUCTION

Odontogenic carcinomas are malignant odontogenic epithelial tumors that are of odontogenic origin and that bear little or no resemblance to odontogenic apparatus.[1] Shafer initially put forth the terminology of Ameloblastic carcinoma in 1974.[2] Previously it was accepted and described as a malignant counterpart of ameloblastoma but according to WHO in 2022, it is considered as a primary odontogenic carcinoma histologically resembling ameloblastoma.[3] Here we report a case of ameloblastic carcinoma of the anterior mandible in a 75-year-old patient, ruling out few troublesome differential diagnosis.

CASE REPORT

A 75-year-old male patient reported with the chief complaint of swelling in the lower front region of jaw for the past 1 year to a private dental hospital at Madurai. The patient initially had minor swelling, but it expanded over time, making it harder for him to close their mouth and masticate. He also occasionally experienced pain and mandibular paresthesia.(Fig:1)

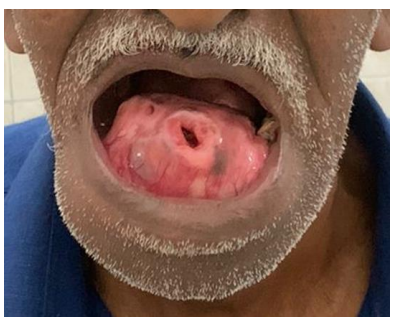

FIG 1: Intra-oral photograph showing bizarre swelling in the anterior edentulous mandible

On intra-oral examination, a bizarre swelling of approximately 7x7 cm was seen in the edentulous alveolar ridge covering the symphysis and body of the mandible on both the sides. Presence of an opening at the surface of the swelling resembling extracted socket but there was no extraction history. The surface mucosa was normal in colour. The cervical examination revealed lymphadenopathy.

The CT imaging revealed expansile radiolucency measuring 70x70 mm in the symphysis and body of the mandible without cortical perforation. Extreme bucco-lingual expansion and cortical thinning were evident such that the cortical borders were ill-defined and difficult to appreciate in the imaging.(Fig:2) Based on the clinical and radiographical features, the differential diagnosis were ameloblastoma and any type of odontogenic carcinomas.

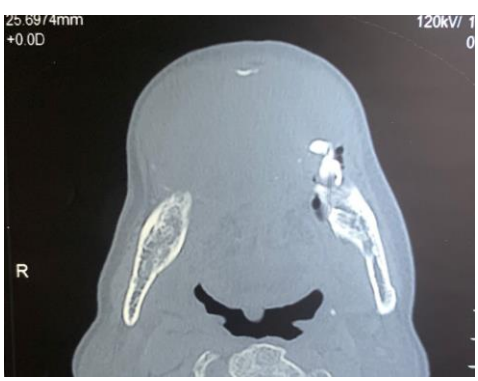

FIG 2: Ill-defined radiolucency with cortical expansion and thinning

Keeping the aggressiveness of the lesion in concern, rather than going for an incisional biopsy, the treatment plan involved segmental mandibulectomy of the lesional area of the anterior mandible followed by reconstruction. The necessity for the surgical procedure, the chosen method as well as potential post-operative risks and complications were thoroughly discussed with the patient. After the detailed conversation , the patient consented to the proposed treatment plan and the resection was done under general anesthesia. The resected mandible was sent for histopathological examination to the Department of Oral and Maxillofacial Pathology, Best Dental Science College, Madurai, for ensuring a thorough evaluation and precise diagnosis.(Fig:3)

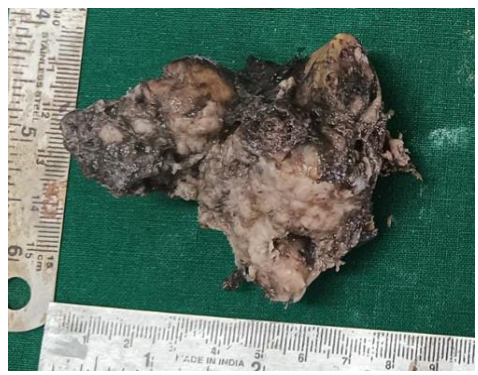

FIG 3: Grossing image of resected mandible

Microscopically, it showed nests and cords of tumor islands resembling ameloblastoma-like component (plexiform pattern).(Fig:4) The epithelium showed peripherally palisaded tall columnar cells resembling ameloblasts and hypercellular stroma. The tumor epithelium showed cellular atypia such as basilar hyperplasia, pale and vesicular nuclei, nuclear and cellular pleomorphism and nuclear hyperchromatism with condensed stellate reticulum. Mitotic activity was not evident.(Fig:5)

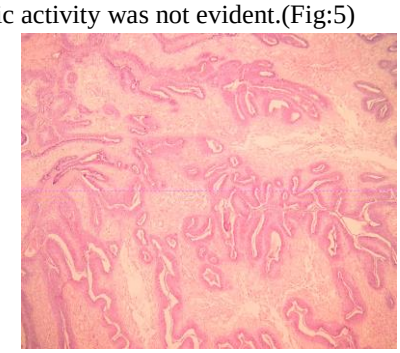

FIG 4: Nests and cords of ameloblastoma like epithelium in hypercellular stroma exhibiting plexiform pattern

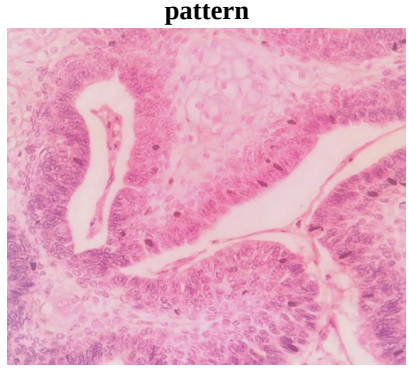

FIG 5: Tumor epithelium showing cellular atypia

Based on the microscopic evidences, the differential diagnosis included Ameloblastic carcinoma, Clear Cell Odontogenic Carcinoma(CCOC), Basaloid Squamous Cell Carcinoma(BSCC) and Primary IntraOsseous Carcinoma(PIOC). Presence of ameloblastoma like component helped to exclude PIOC from the option. Periodic Acid Schiff (PAS) staining was conducted, revealing negative expression of mucin in the microcystic areas,(Fig:6) thus eliminating the possibilities of CCOC and BSCC. Immunohistochemistry (IHC) demonstrated consistent staining for Ki-67, indicating a higher rate of cell proliferation in the highlighted regions.(Fig:7) Therefore, based on the above evidences, the definitive final diagnosis of Ameloblastic carcinoma was established.

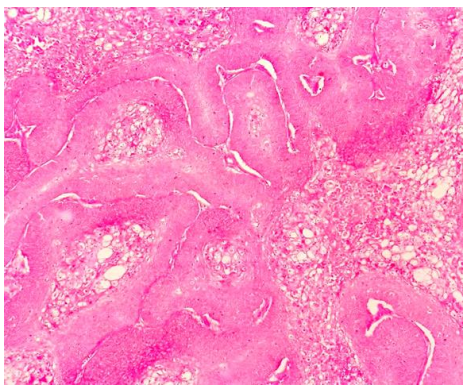

FIG 6: Negative expression for mucin in PAS stain

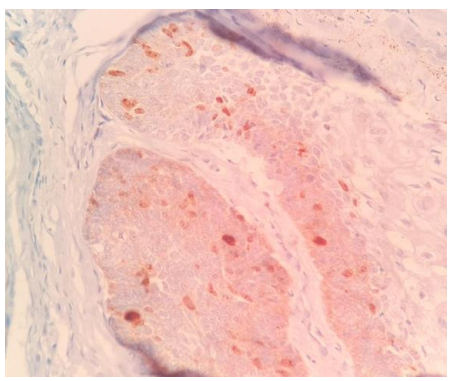

FIG 7: Ki-67 Labelling Index showing positivity in highly proliferative areas.

At the five-month follow up, there was no evidence of recurrences and rehabilitation for the patient will be scheduled in the near future.

DISCUSSION

Odontogenic malignancies are rare and comprise about 1% of all jaw tumors. AMCa is a uncommon, aggressive malignant epithelial odontogenic tumor of the jaw with an incidence of less than 1%, with 28% of the reported cases in India.[4] AMCa was acknowledged by the WHO as a distinct entity in 2005. It is characterized as a tumor with ameloblastomatous differentiation showing cytologic features of malignancy with or without metastasis. This was changed in 2022 as a primary odontogenic carcinoma histologically resembling ameloblastoma.[3]

Men are more commonly affected by AMCa and it typically affects the posterior mandible. It is categorized into three types: primary, secondary-intraosseous and secondary-peripheral.[2,4]

Most of the cases arise spontaneously (*denovo*) with few cases arising from long-standing or recurrent ameloblastoma and odontogenic cyst. BRAF p.V600E mutations have been reported in AMCa, but it has no defined diagnostic value.[3]

The clinical symptoms are more aggressive with rapid growth, cortical expansion, perforation, facial asymmetry, deformity, pain and paraesthesia. Radiologically, it presents as unilocular or multilocular radiolucency or mixed radioopacities with ill-defined borders.[4] Hall *et al* suggests that there are four clinical criteria that can aid in diagnosing ameloblastic carcinoma. These include rapid growth, tendency to perforate the cortex, pain, and paresthesia, which are different from those seen in the benign counterpart.[5]

Microscopically, it is composed of islands and cords of ameloblastoma-like component, predominantly plexiform and follicular pattern. The tumor epithelium shows features of cellular atypia with condensed stellate reticulum in a hypercellular stroma. Clear cell differentiation are rare. SOX-2 has recently been shown to be a sensitive and specific IHC marker for AMCa that is negative in ameloblastoma.[6]

In a previous case report [7], the tumor was found to be in the anterior mandible involving symphyseal region which is identical to this case report. In contrast to this case, necrosis and mitotic activity was found to be the common findings in most of the cases in the literature. In another case presentation of AMCa [8], Ki-67 LI was found to be almost 20% in the areas of increased proliferation, which was similar to our findings.

Metastasis is most common especially to lungs with a recurrence rate of 20.9-38.4%, which necessitates long term follow-up. Prognosis depends on the aggressiveness, location, local destruction and distant metastasis. The 5-year-survival rate is higher (69.1-83.2%), but decreases in metastatic cases. The treatment of choice is wide surgical resection with 2-3cm of bony margins with or without lymph node dissection. Adjuvant radiotherapy and chemotherapy are beneficial.[2,4,7]

CONCLUSION

Diagnostic dilemma is common among odontogenic tumors because of the similar clinical, radiographical and histopathological features. This case report highlights a challenging rare case of ameloblastic carcinoma in the anterior mandible with multiple diagnostic difficulties.

FINANCIAL SUPPORT AND SPONSORSHIP

Nil

CONFLICTS OF INTEREST

There are no conflicts of interest

ACKNOWLEDGMENTS

We would like to express our gratitude to our patient for his co-operation and consent, which allowed us to share his clinical information and images for the benefit of medical knowledge. Also, we are grateful to Dr.C.S.Prabahar,M.D.S., Principal, Best Dental Science College and the following faculty of the Department of Oral and Maxillofacial Pathology, Dr.G.Kesavan M.D.S.,(Reader) and Dr.S.Soundarya M.D.S., (Reader) for their valuable inputs and Mrs.K.Manimegalai (Lab technician) for her support.

PATIENT'S CONSENT

We assure that we have obtained the necessary patient consent using the patient consent form from the patient involved in this case report. Within this consent form, the patient has granted permission for disclosing his data and images for the purpose of advancing medical knowledge. The patient is well aware that his personal identity, including his name and initials, will remain confidential, and all necessary measures will be taken to ensure his anonymity.

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