MEDICAL SCIENCE

To Cite:
Kumar KS, Giri GVV, Kumar JN, Vishal R, Madhumitha R. Incidentaloma: Ameloblastic carcinoma of mandible: A case report. *Medical Science* 2023; 27: e223ms2868.
doi: https://doi.org/10.54905/disssi/v27i135/e223ms2868

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Peer-Review History
Received: 26 January 2023
Reviewed & Revised: 30/January/2023 to 04/May/2023
Accepted: 08 May 2023
Published: 12 May 2023

Peer-review Method
External peer-review was done through double-blind method.

Medical Science
pISSN 2321–7359; eISSN 2321–7367

Incidentaloma: Ameloblastic carcinoma of mandible: A case report

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ABSTRACT

Incidentaloma is a radiological neology for incidental findings unrelated to the original diagnostic inquiry which helps in early diagnosis of suspicious lesions. Ameloblastic carcinoma is an uncommon and aggressive odontogenic tumour which causes significant bone destruction and shows cytologic characteristics of malignancy. It's more commonly seen in the mandible and in wide range of age groups. This case report aims at presenting an incidental finding of ameloblastic carcinoma in an asymptomatic patient who sustained facial fractures following trauma.

Keywords: Malignant ameloblastoma, benign tumour of mandible, Incidentaloma.

1. INTRODUCTION

Incidentaloma is a radiological coinage for an incidental finding which an unanticipated finding is not related to original diagnostic inquiry. An incidental imaging finding can help in early diagnosis of some dubious lesions of clinical significance in some cases, whereas in other cases it may cause overdiagnosis and result in a cascade effect causing more problems to the patient (Lumbreras et al., 1983). According to WHO definition, ameloblastic carcinoma is defined as a rare primary odontogenic malignancy that mixes the histological features of ameloblastoma with cytological atypia (Kramer et al., 1992). The incidence of ameloblastic carcinoma reported within the literature is rare and majority of ameloblastic carcinomas involve the mandible and only few cases have reported to occur within the maxilla. There is no gender predilection and is seen to occur in both sexes. The posterior segments of the mandible represent the foremost common site. Rapidly progressing painful swelling is commonly seen and it could also resemble a cyst like lesion having benign clinical features or as a tissue mass that has outsized with ulceration. Findings of bone resorption and tooth mobility are also seen. Cortical expansion often with perforation could even be present likewise as infiltration into adjacent structures (Lolachi et al., 1995; Ozlugedik et al., 2005; Avon et al., 2003).

Malignant ameloblastoma and ameloblastic carcinoma are used interchangeably although different; the former tends to metastasize even

though it is a benign tumour while ameloblastic carcinoma shows features of both ameloblastoma and carcinoma (Slootweg and Müller, 1984; Corio et al., 1984; Elzay, 1982). However, the treatment of ameloblastic carcinoma is controversial as no consensus has been obtained about the treatment. Typically followed treatment modality is wide surgical excision with or without radiotherapy. Here we present a case of ameloblastic carcinoma which was an incidental finding when patient was evaluated for facial fractures.

2. CASE PRESENTATION

A 54-year-old male patient reported to OMFS OPD of Sri Ramachandra Dental College and Hospital with primary complaint of pain in relation to mandible following a road traffic accident, skid and fall from two-wheeler and had sustained injury to face. The patient was neurologically stable. On local examination, patients face was apparently symmetrical; there was presence of tenderness in relation to left parasymphysis region and right condyle and angle region of mandible. Mobility of teeth was evident in relation to lower right third molar. Mouth opening was adequate (Figure 1).

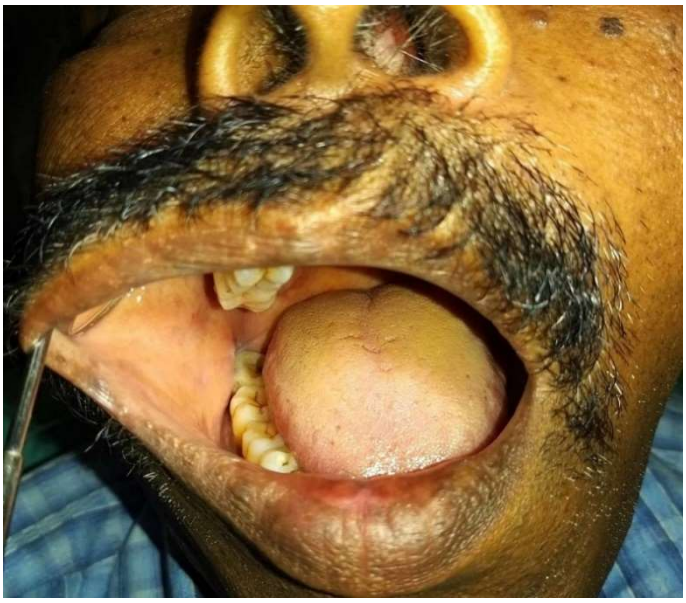

Figure 1 Intraoral clinical presentation of the patient (preoperative)

Preoperatively radiological investigation (CT scan) revealed presence of a pathologic fracture of right condyle of mandible and a radiolucent lesion of size 6cmx2cmx2cm in relation to right posterior mandible extending from distal aspect of lower right second molar to neck of right condyle and an undisplaced fracture of left parasymphysis of mandible was also noted (Figure 2A, 2B, 3A, 3B).

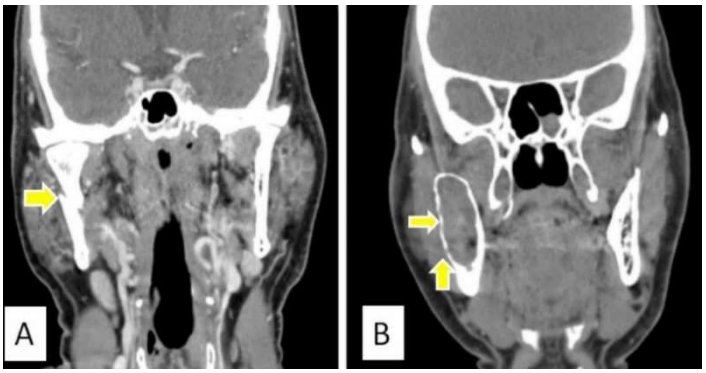

Figure 2 A: Coronal section of CT showing pathologic fracture of condyle; B: Coronal section of CT displaying radiolucent lesion in relation to right posterior mandible

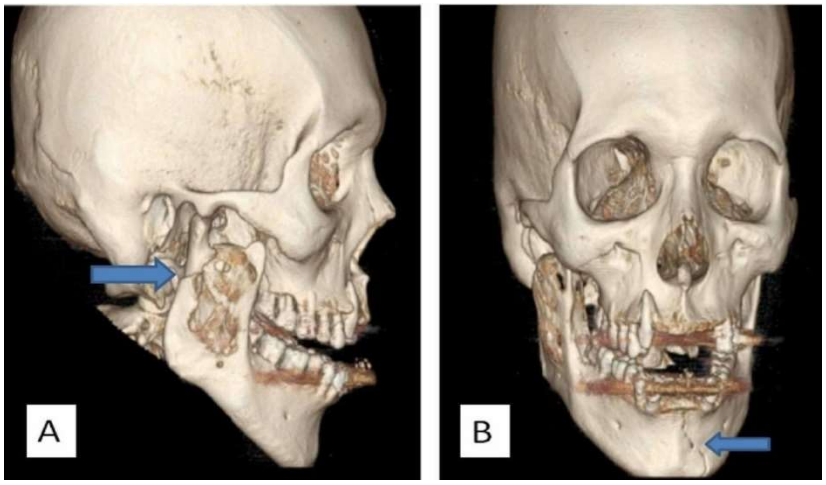

Figure 3 A: 3D image of CT showing bony lesion invading right posterior mandible; B: 3D image of CT showing undisplaced fracture of left parasymphysis of mandible

An incisional biopsy done under local anaesthesia revealed ameloblastic carcinoma of right mandible. All baseline investigations were done and it was in the normal range. Patient was treated by right hemi mandibulectomy involving right condyle sparing the lower anterior teeth and the masseter, medial pterygoid and lateral pterygoid muscles were removed for surgical margin clearance (Figure 4).

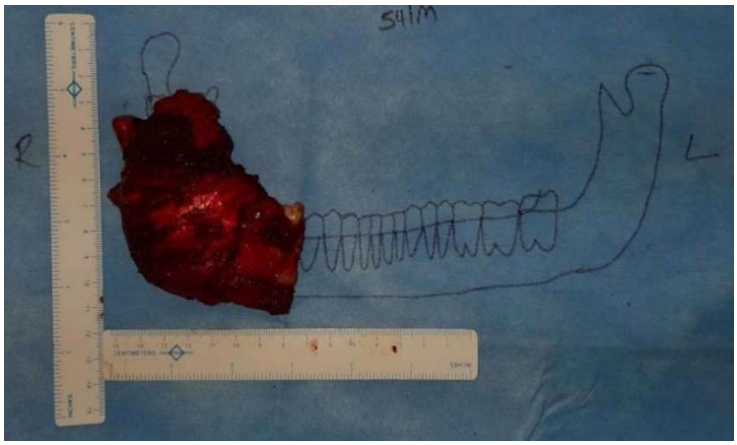

Figure 4 Resected mandible specimen

Level Ib lymph nodes were sent for frozen section and it had been found to be negative for malignancy. The resected specimen was sent for histopathologic examination. The surgical defect in relation to right posterior mandible was reconstructed using pectoralis major myocutaneous flap.

The final histopathology report suggested the margins were free from tumour and it revealed areas of necrosis with focal areas of odontogenic epithelial islands arranged as follicles and strands within the tissue stroma lined by tall columnar cells enclosing stellate reticulum like cells with features of malignancy with cellular pleomorphism, nuclear hyperchromatism and increased nuclear cytoplasmic ratio with areas of keratin pearl formation and inductive dentinoid formation and peripheral stratified squamous epithelium was seen (Figure 5). Patient was followed up one year post operatively and no recurrence was noted (Figure 6).

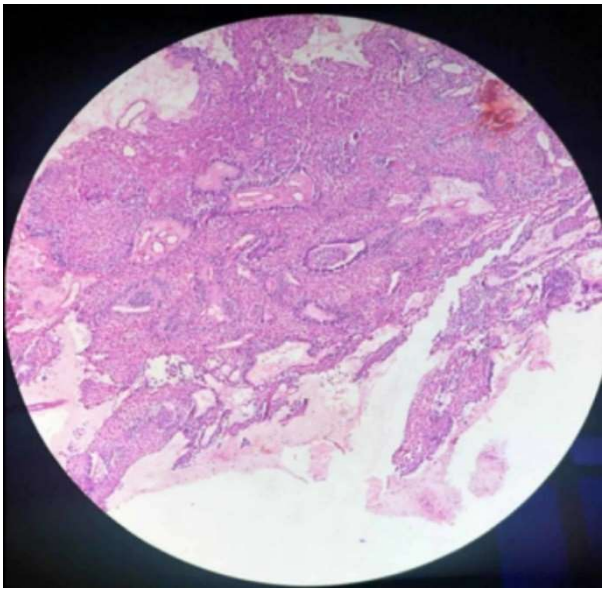

Figure 5 Histopathologic section

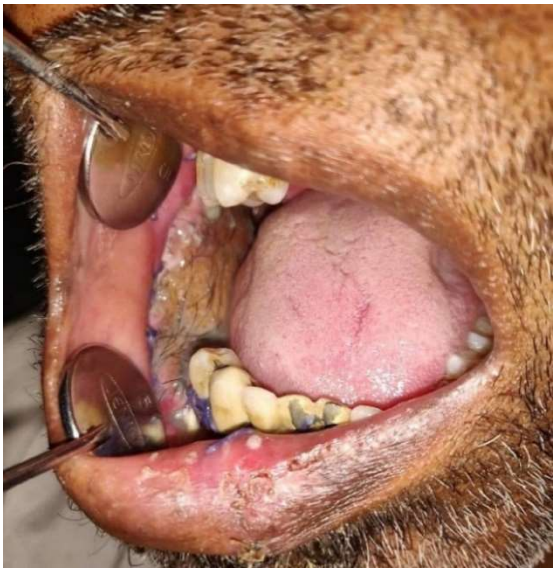

Figure 6 Intraoral photograph displaying healthy flap uptake (one month post-operative)

3. DISCUSSION

Lumbreras et al., (1983) performed a scientific review on the incidental findings in imaging diagnostics on frequency and management of those findings. From the 26 included CT articles they found the mean frequency of incidental findings was 31.1% (Lumbreras et al., 1983). Only one study has classified the incidentalomas into groups based on their potential clinical significance and located that 16.1% required follow-up or referral. Only one study on incidentalomas had reported among a sample size of 1000 subjects, in that incidental finding of three malignancies were found (Allareddy et al., 2012).

Ameloblastic carcinoma is an uncommon malignant odontogenic epithelial neoplasm that arises de novo or from a pre-existing odontogenic lesion. In between 1984 to 2012, about 92 cases have been reported in scientific literature. Males are commonly affected, most commonly affecting the mandible (Ozlugedik et al., 2005). The clinical features of ameloblastic carcinoma include rapid jaw expansion, associated with pain and often cause cortex perforation.

In few cases of ameloblastic carcinoma, regional and distant metastasis is seen although it is a feature of malignant ameloblastoma. Metastasis to the lung or regional lymph nodes is sometimes seen (Eversole, 1995). Mac-Intosh, (1991) reported that ameloblastic metastasis was first reported in the lung which was thought to be aspirated from oral lesion due to curettage and enucleation surgeries which might liberate neoplastic cells into the upper airway spreading to lower airway. Ameloblastoma may undergo malignant transformation spontaneously following radiation or following chemotherapy (Reichart et al., 1995; Gardner,

1996). In two of its variants - granular cell type and also clear cell type, aggressive behaviour and metastatic potential is seen. Expression of parenchymal matrix metalloproteinases2 (MMP2), cytokeratin 18, Ki67, stromal MMP9 and Ki67 was compared in a study by Yoon et al., (2011) which differentiated ameloblastic carcinoma from ameloblastoma.

Literature suggests only few case reports in which neck was addressed. In our case we did a sentinel node biopsy and it had been negative and further neck dissection wasn't done. There is still controversy regarding neck dissection for Ameloblastic Carcinoma in clinically and radiologically negative neck. Surgical resection is the gold standard treatment for ameloblastic carcinoma. In order to confirm a disease-free survival an en bloc resection with 1-2 cm of normal bone margin the safest surgical modality which has shown local recurrence rates of about 15%. Atkinson et al., (1984) described the effect of megadose radiation in 10 patients with ameloblastoma out of which nine patients responded and three among them underwent surgical salvage. Seven of the ten patients, showed no evidence of disease post-surgery and/or radiation, with follow up ranging from one to ten years (Atkinson et al., 1984).

Gardener et al recommended a radiation dose between 3,000 cGy -5,000 cGy (Gardner, 1988) Ameloblastic carcinomas is predominantly intraosseous. Therefore, the effectiveness of radiation treatment should be considered. Ramadas et al., (1990) found the use of Adriamycin, cyclophosphamide, methotrexate, leucovorin and cisplatin to be beneficial (Ramadas et al., 1990). The survival rate is majorly determined by regional or distant metastasis and local recurrence. Infante-Cossio et al., (1998) published that in two of the patients who had a maximum five years of follow up had no evidence of local recurrence or metastasis. According to Infante-Cossio et al., (1998) five years of survival rate after surgery and radiotherapy without recurrence or metastasis is accepted.

4. CONCLUSIONS

This case report gives an insight about ameloblastic carcinoma, which has very limited evidence in the literature and emphasises on the importance of routine radiological examination which has to be included in the regular dental check up to diagnose such pathologies in earlier stage as most of the pathologies in early stages are asymptomatic and may only be identified incidentally in radio graphical examination. Ameloblastic carcinoma cases must be studied carefully histologically in order to differentiate it from ameloblastoma and malignant ameloblastoma with proper assessment of nodal metastasis. Most accepted treatment modality remains surgical resection and may require neck dissection if nodal involvement is present. The role of radiotherapy and chemotherapy still stands controversial.

Author Contributions

Dr K Santhosh Kumar: Operating surgeon, selection of case, manuscript preparation and data analysis
Dr G V V Giri: Operating surgeon, manuscript preparation, data analysis and figures selection.
Dr J Naveen Kumar: Operating surgeon, manuscript review, data analysis and interpretation.
Dr Vishal R: Manuscript preparation and editing.
Dr Madhumitha M: Manuscript preparation and editing.

Abbreviations

WHO – World Health Organisation
OMFS – Oral and Maxillofacial Surgery
OPD – Outpatient Department
CT – Computed Tomography

Acknowledgements

Nil

Informed consent

Written & oral informed consent was obtained from the individual participant included in the case report.

Funding

This study has not received any external funding.

Conflict of interest

The authors declare that there is no conflict of interests.

Data and materials availability

All data sets collected during this study are available upon reasonable request from the corresponding author.

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