Case Report

Ameloblastic Carcinoma: Presentation of a Rare Case

Palak H Shah, Rashmi Venkatesh, Chandramani B More, Vassandacoumara Vaishnavee

Department of Oral Medicine and Radiology, Kanjibhai Manjibhai Shah Dental College and Hospital, Sumandeep Vidyapeeth University, Vadodara, Gujarat, India

Abstract

Ameloblastic carcinoma is a rare malignant tumor of odontogenic origin. It is an aggressive neoplasm that is locally invasive and can spread to regional lymph nodes or distant sites such as lung and bones. Histopathology is a vital tool for diagnosis of ameloblastic carcinoma and is also required to differentiate it from ameloblastoma. Its biologic behavior recommends necessity of early diagnosis and prompt treatment. However, due to lack of case reports with long-term follow-up, there is no established treatment protocol for these cases. Here, we are presenting a unique case of ameloblastic carcinoma of left mandible in a 48-year-old female patient treated successfully with resection and reconstruction by fibular graft.

Keywords: Ameloblastic carcinoma, odontogenic carcinoma, odontogenic tumor

Introduction

Ameloblastoma is a benign tumor of the odontogenic epithelium with 36.9% occurrence among all the odontogenic tumors.[1] The malignant transformation of ameloblastoma is a matter of debate since years and two variants have been identified – malignant ameloblastoma and ameloblastic carcinoma. The extremely rare occurrence of these lesions explains the lack of detailed description about the characteristics of these entities. Slootweg and Muller (1984) and Corio *et al*. (1987) defined ameloblastic carcinoma as any ameloblastoma with histologic evidence of malignant disease in primary or recurrent tumor, regardless of metastasis.[2] Ameloblastic carcinoma is a rare malignant odontogenic neoplasm characterized by aggressive behavior and poor prognosis. The occurrence ratio of ameloblastic carcinoma to malignant ameloblastoma is 2:1.[3]

Case Report

A 48-year-old female patient reported to the Department of Oral Medicine and Radiology, Kanjibhai Manjibhai Shah Dental College and Hospital, Sumandeep Vidyapeeth University, Piparia, Vadodara, Gujarat, India with the chief complaint of an extraoral swelling on left side of the face since 3 years. The swelling was initially small in size and slowly increased and attained the present size. As the swelling was asymptomatic, the patient completely ignored it. During the course of the swelling, the teeth in the region became mobile and exfoliated but still the patient ignored it as it was asymptomatic. Patient was cachectic with history of weight loss. On extraoral examination, a single well-defined swelling [Figure 1] was present on lower third of the left side of face causing facial asymmetry. The swelling was roughly oval in shape and approximately 3 × 4 cm in size. The overlying skin appeared stretched and shiny. On palpation, the swelling was hard and non-tender. Paresthesia was elicited in the left mandibular region extraorally.

On intraoral examination, the left mandibular quadrant was completely edentulous. Buccal vestibular obliteration was present in the same region with proliferative growth covered by slough. The growth was extending from premolar region to 3rd molar region anteroposteriorly. On palpation, the growth was nontender and firm in consistency. Expansion of lingual cortical plate was felt with discontinuation of cortical plate in the premolar region [Figure 2]. Based on the history and

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Address for correspondence: Dr. Palak H Shah, Department of Oral Medicine and Radiology, Kanjibhai Manjibhai Shah Dental College and Hospital, Sumandeep Vidyapeeth University, Piparia, Vadodara, Gujarat, India.
E-mail: shahpalak98@gmail.com

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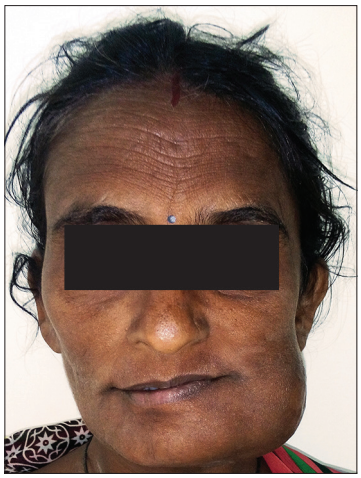

Figure 1: Extraoral swelling on lower 1/3rd of left side of face

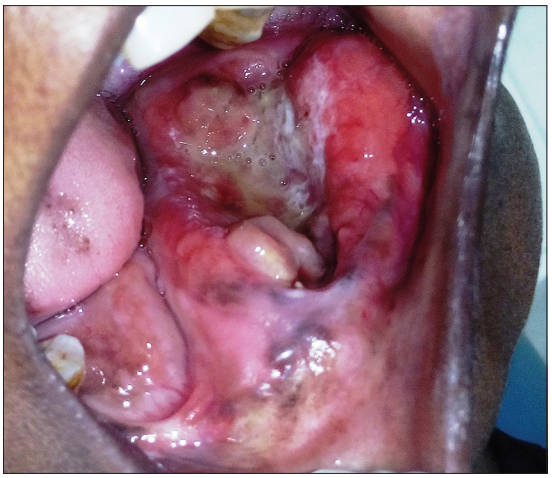

Figure 2: Intraoral ulceroproliferative growth covered by slough

clinical findings, a provisional diagnosis of ameloblastoma was made and odontogenic myxoma was included as a differential diagnosis. Due to the cortical plate disruption, the possibility of ameloblastic carcinoma or primary intraosseous carcinoma was also considered for the differential diagnosis.

On radiographic investigation, panoramic radiograph revealed [Figure 3] a large, multilocular lesion involving left side of mandible extending anteriorly from symphysis menti and involving ramus posteriorly. There was expansion of the lower border of mandible. Ultrasonography [Figure 4] showed a mixed hyper and hypoechoic lesion. Plain computed tomography (CT) scan [Figure 5] showed an expansile lobular lesion with multiple septations in the mandibular body and ramus region with thinning of the cortex. There was no evidence of calcification. The CT report stated that the lesion was extending to the left temporomandibular joint and inferior temporal fossa superiorly, close to the alveolar process inferomedially, to the cheek antero-laterally, and posteriorly it had a retro-oral extension. Post-contrast CT [Figure 6] showed irregular enhancement of the tumor. On radiographic investigations, the radiographic diagnosis of ameloblastoma was made and possibility of primary intraosseous carcinoma was ruled out. However, a possibility of ameloblastic carcinoma could not be ruled out.

After radiographic investigations, hemogram was obtained and incisional biopsy was performed. Histopathological examination revealed connective tissue stroma and epithelial islands along with follicles. Connective tissue was predominantly cellular, infiltrated with the follicles, islands, strands, and cords of epithelial cells. Basal cells (lining cells) of some of the follicles were tall columnar in shape showing reversal of polarity. All features resembled ameloblast-like cells and odontogenic epithelium. Connective tissue was also infiltrated with the inflammatory cells chiefly comprising of chronic inflammatory cells. At one focal area, connective tissue was infiltrated with the cords and many single scattered epithelial cells were present showing exceedingly dysplastic like features, extensive cellular and nuclear pleomorphism and an enormous number of abnormal mitotic figures [Figure 7].

With the clinicoradiographic and histopathological examinations, the diagnosis of ameloblastic carcinoma was given. The patient was treated surgically in the Department of Oral and Maxillofacial Surgery where hemimandibulectomy [Figure 8] followed by microvascular reconstruction with fibular graft was performed. The postoperative period was uneventful.

Discussion

Ameloblastic carcinoma is a rare malignant tumor of odontogenic origin with only 92 cases reported in the scientific literature during 1984–2012.[4] The tumor shows an age distribution of 5th to 7th decade of life with a male predilection.[5] Our case demonstrated ameloblastic carcinoma occurring in a middle-aged woman. Mandible is more commonly seen to be involved as compared to maxilla.[6] Similarly, our case also demonstrated involvement of the mandible.

The ameloblastic carcinoma can develop in cases of long-standing ameloblastomas. Once developed, it shows more aggressive behavior. The most common clinical presentation of ameloblastic carcinoma is a facial swelling with marked perforation of cortical plates and extension into adjacent soft tissues.[7] Our case exhibited similar presentation. The radiographic appearance of ameloblastic carcinoma is consistent with that of ameloblastoma except for occasional presence of some focal radiopacities, apparently reflecting dystrophic calcification.[8] In our case, no calcification was noted on radiographic examination.

WHO (2005)[9] has classified malignant tumors of odontogenic origin. According to which, carcinoma associated with ameloblastoma can be classified as metastasizing (malignant) ameloblastoma or ameloblastic carcinoma. Metastasizing ameloblastoma is the one which shows distant metastasis and the primary tumor demonstrates benign histological features, whereas, ameloblastic carcinoma is described as a

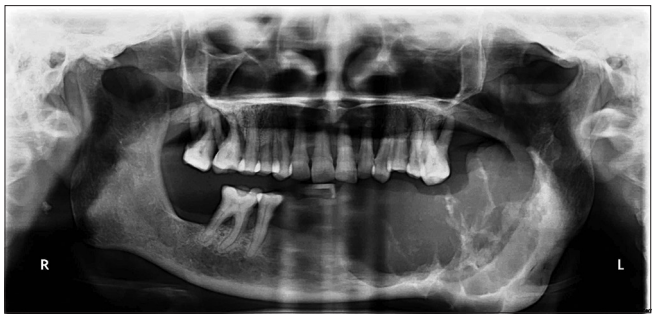

Figure 3: Panoramic radiograph showing an extensive multilocular lesion in left mandible

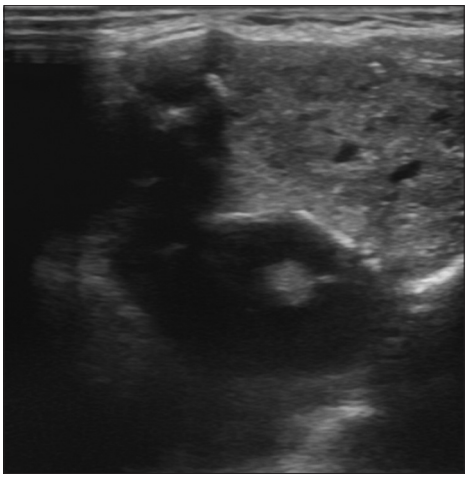

Figure 4: Ultrasonography showing a mixed hyperechoic–hypoechoic lesion

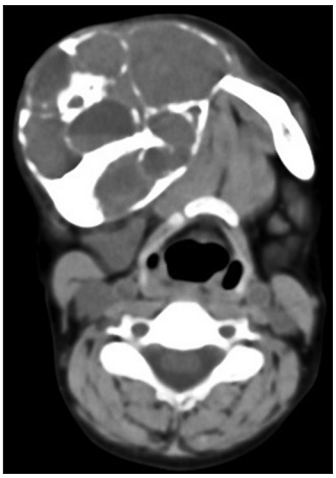

Figure 5: Axial section of plain CT showing expansile lobulated septated lesion

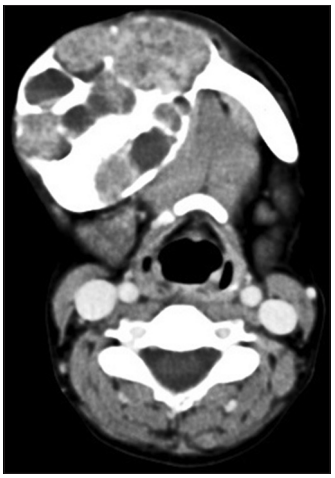

Figure 6: Axial Section of contrast CT showing irregular enhancement of tumor

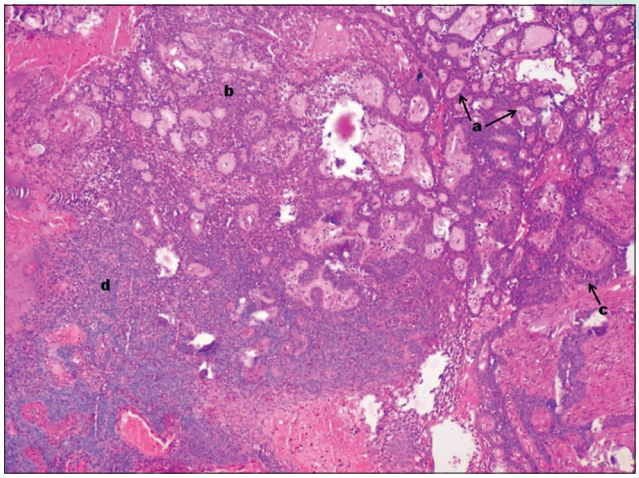

Figure 7: Photomicrograph (4×) (a) Epithelial islands along with follicles; (b) Predominantly cellular connective tissue infiltrated with islands and cords of epithelium; (c) Ameloblast-like cells showing reverse polarity; (d) Focal infiltration of epithelial cells showing dysplastic features

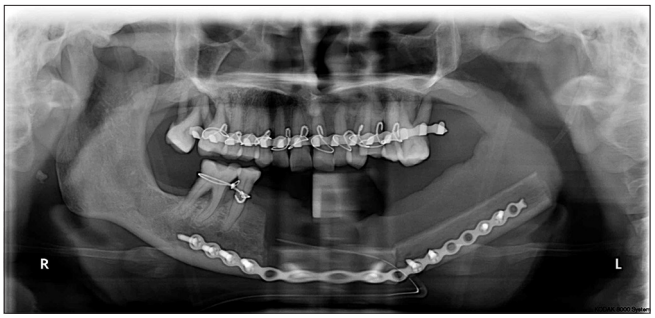

Figure 8: Postoperative panoramic radiograph

tumor that exhibits malignant features along with the features of ameloblastoma with or without metastasis. Ameloblastic carcinoma can be of primary or secondary type. The primary type demonstrates malignancy in the primary tumor. The secondary type consists of malignant changes, which originate in a previously existing ameloblastoma. Our case was of primary type of ameloblastic carcinoma.

In cases of ameloblastic carcinoma, the treatment of choice is extended surgical resection with at least 2-3 cm of bone margins in order to prevent recurrence. This results in recurrence rate of less than 15%.[10] The utility of radiotherapy in cases of ameloblastoma is debatable. Distance metastasis is reported

in cases of ameloblastic carcinoma to the lungs, brain, bones and liver which is a serious condition and at times fatal.[5] In our case, the patient did not demonstrate any clinical feature suggestive of distant metastasis and no radiographic evidence of recurrence was noted after one year of surgery. Further investigations to rule out distant metastasis were advised.

Conclusion

Ameloblastoma demonstrates a spectrum of histologic and biologic behavior ranging from benign to malignant. Therefore, thorough histopathological examination becomes an inevitable step in management of these cases in order to determine their nature and also to guide the treatment plan. Cases showing carcinomatous transformation require prompt surgery and a long-term follow-up.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. Johnson NR, Gannon OM, Savage NW, Batstone MD. Frequency of odontogenic cysts and tumors: A systematic review. J Investig Clin Dent 2014;5:9-14.
2. Langlais RP, Langland OE, Nortje CJ. Diagnostic Imaging of Jaws. Malvern, USA: Williams & Wilkins; 1995. p. 406.
3. Regezi JA, Kerr DA, Courtney RM. Odontogenic tumors: Analysis of 706 cases. J Oral Surg 1978;36:771-8.
4. Kar IB, Subramanyam RV, Mishra N, Singh AK. Ameloblastic carcinoma: A clinicopathologic dilemma: Report of two cases with total review of literature from 1984 to 2012. Ann Maxillofac Surg 2014;4:70-7.
5. Kallianpur S, Jadwani S, Misra B, Sudheendra US. Ameloblastic carcinoma of mandible: Report of case and review. J Oral Maxillofac Pathol 2014;18:96-102.
6. Li J, Du H, Li P, Zhang J, Tian W, Tang W. Ameloblastic carcinoma: An analysis of 12 cases with review of literature. Oncol Lett 2014;8:914-20.
7. Avon SL, McComb J, Clokie C. Ameloblastic carcinoma: Case report and literature review. J Can Dent Assoc 2003;69:573-6.
8. Naik V, Kale AD. Ameloblastic carcinoma: A case report. Quintessence Int 2007;38:873-9.
9. Barnes L, Eveson J, Reichart P, Sidransky D. World Health Organization classification of tumours; pathology and genetics of head and neck tumors. IARC Press 2005.
10. Kishore M, Panat SR, Aggarwal A, Upadhyay N, Agarwal N. Ameloblastic carcinoma: A rare case report. J Clin Diag Res 2015;9:ZD27-8.