A Rare Condition of Ameloblastoma Transforming into Ameloblastomic Carcinoma - A Case Report

Manojnya Somabhatta, Jakkula Ananthnag, Srikanth Damera, V. R. Chandrababu Pamidi, Tarun Gogineni[1], Sriram Burugupalli[1]

Department of Oral and Maxillofacial Surgery, GSL Dental College and Hospital, [1]GSL Cancer Trust, Rajahmundry, Andhra Pradesh, India

Abstract

Rationale: Ameloblastic carcinoma is defined as an ameloblastoma in which there is histological evidence of malignancy in primary tumour or recurrent tumour regardless if it has metastasised or not. It is aggressive in nature. **Patient Concerns:** The patient presented with a painful swelling associated with restricted mouth opening. **Diagnosis:** This is the clinical profile of a patient who has presented with a recurrent ameloblastic carcinoma. **Treatment:** Histological features of the excised specimen resemble ameloblastic carcinoma constituting cytological atypia with tall columnar cells. **Outcome:** Excision of tumour was done. Reconstruction was done using pectoralis major myocutaneous flap. **Take-away Lessons:** Ameloblastic carcinoma is an aggressive tumour and constitutes destruction and distant metastatic spread. Hence, aggressive resection is the choice of treatment along with a long-term follow-up for better quality of life.

Keywords: Ameloblastic carcinoma, ameloblastoma, cytological atypia, recurrence

Introduction

Ameloblastoma is a benign, locally aggressive odontogenic neoplasm, causing slowly growing painless swelling of jaws. It constitutes about 1–3% of all jaw tumours and cysts. The maxillomandibular ratio is 5:1[1] and the most common site of occurrence is the mandibular molar region.[1] Ameloblastic carcinoma is used to refer to any ameloblastoma with histologic evidence of malignancy in the primary tumour or recurrent tumour regardless of metastasis.[2] It constitutes 1.5–2.0% of all odontogenic tumours, arising as a new lesion or in a pre-existing ameloblastoma. In 1982, Elzay[3] described ameloblastic carcinoma as a malignant epithelial odontogenic tumour which histologically retains the features of ameloblastoma. Here, we present a case of ameloblastic carcinoma of mandible.

Case Report

A 37-year-old female reported to the Department of Oral and Maxillofacial Surgery, with a chief complaint of swelling in the right lower third of the face for the past eight months. A history of swelling associated with continuous, moderate, dull, progressive and throbbing pain along with painful and restricted mouth opening was noted [Figure 1]. The patient was under medication for seizures for 10 years. She was hepatitis B positive, with a history of surgery four years back for a swelling which was then diagnosed as ameloblastoma and a reconstruction plate was placed to stabilise the mandible. The patient was subjected to routine general examination. On extraoral examination, a diffuse solitary swelling of size 6 x 4 cm extending from infraorbital ridge to lower border of mandible supero-inferiorly and nasolabial sulcus to tragus of right ear antero-posteriorly. The swelling was firm in consistency with tenderness on palpation and no local rise of temperature. On intraoral examination, the swelling was in the right buccal vestibule and was smooth, diffuse, solitary and firm in consistency with ill-defined margins.

On contrast-enhanced computed tomography (CECT) of the head and neck, a large fairly well-defined enhancing

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Address for correspondence: Dr. Manojnya Somabhatta, Department of Oral and Maxillofacial Surgery, GSL Dental College and Hospital, Rajahmundry, Andhra Pradesh, India.
E-mail: burramanojnya@gmail.com

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lesion measuring 7.6 cm × 6.8 cm × 5.4 cm was noted involving the right upper and lower buccal mucosa, right masticatory muscles and abutting anteriorly the subcutaneous fat and posteriorly the parotid gland and the zygomatic arch and medially, the lateral pterygoid plate, right submandibular gland, lateral aspect of tongue and hard palate superiorly [Figure 2].

Apron incision was given on the right side for the neck dissection. Neck dissection was done involving the level 1, 2 and 3 lymph nodes. This incision was continued as lip split incision of the lower lip extending from labial and lingual vestibule superiorly exposing the condyle. Excision of lesion, disarticulation of mandible, removal of previously placed reconstruction plate followed by partial maxillectomy was done [Figure 3]. As the maxilla was involved on the posterior surface and pterygomaxillary area, to achieve complete clearance, partial maxillectomy was done using the intraoral approach with resection of posterior teeth and the alveolus [Figure 4]. Reconstruction was done with pectoralis major myocutaneous flap. Drains were secured. Closure was done using 3-0 vicryl and 3-0 silk sutures. The specimens were sent for histopathological examination. The patient was followed up for two years and was advised chest X-ray for follow-up of distant metastasis [Figure 5].

The tissues showed varying sized nests, trabecular glands and islands of columnar epithelial cells, which showed eosinophilic cytoplasm and round to oval hyperchromatic nuclei showing reverse polarisation. The centre of these nests showed squamous differentiation [Figure 6].

Discussion

Odontogenic malignancies are rare and account for 1% of all cysts and tumours of jaws. In 1983, Shafer introduced the word ameloblastic carcinoma to describe ameloblastoma which undergo histologic malignant transformation. Clinically and radiographically, both can be differentiated if there is a sudden increase in size of swelling, pain, expansion and perforation of cortical plate with soft tissue extension or any foci of calcifications. In the present case report, the patient presented with pre-operative swelling, tenderness on palpation along with rapid growth and restricted mouth opening. In the WHO 2005 classification, ameloblastic carcinoma was divided into three categories: primary, secondary and peripheral. These tumours were classified under 'Ameloblastic Carcinoma' in the WHO 2017 classification based on the morphologic features and similar behaviour. According to Dhir *et al*., the age range of appearance of ameloblastic carcinoma was 51–84 years.[4] Posterior mandible was the most common site of occurrence.[2,5]

Hypermethylation of p16 tumour suppressor gene was reported in the malignant transformation of ameloblastoma to ameloblastic carcinoma.[6] The patient was not subjected for immunohistochemistry investigations. Radiologically, ameloblastoma and ameloblastic carcinoma can be unilocular or multilocular with distinct borders in ameloblastoma and ill-defined borders in ameloblastic carcinoma. Loss of lamina dura along with root resorption was noted. It later showed local radiopacity, reflecting dystrophic calcifications. The borders showed marginal sclerosis without periosteal new bone formation.[5]

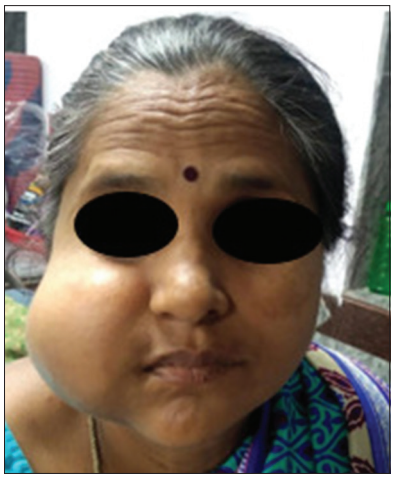

Figure 1: Pre-operative view of patient

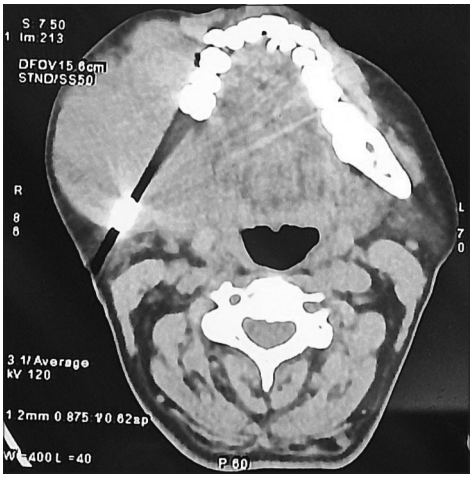

Figure 2: Axial view of contrast enhanced computed tomography showing the lesion on right side of the mandible

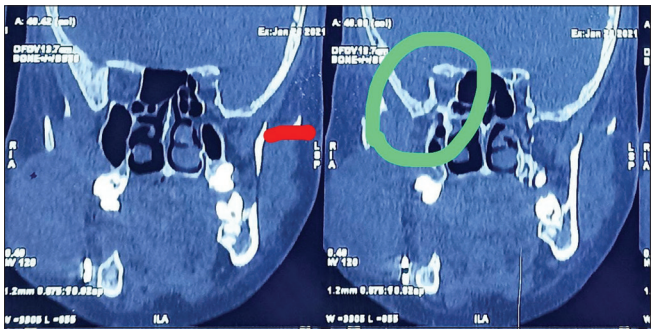

Figure 3: Coronal view of contrast enhanced computed tomography; the lesion invading the pterygomaxillary space – green circle, red line – normal landmarks on left side

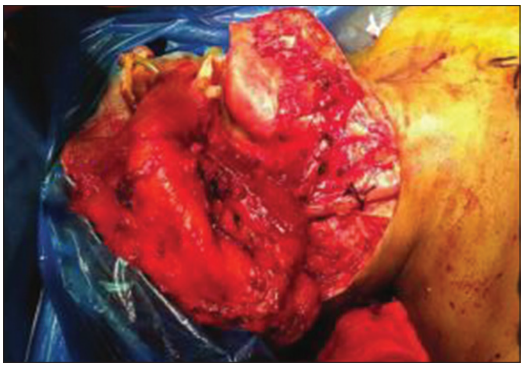

Figure 4: Intraoperative image after resection

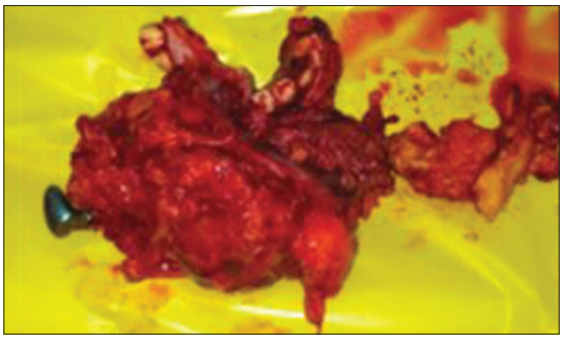

Figure 5: Specimen for histopathological examination

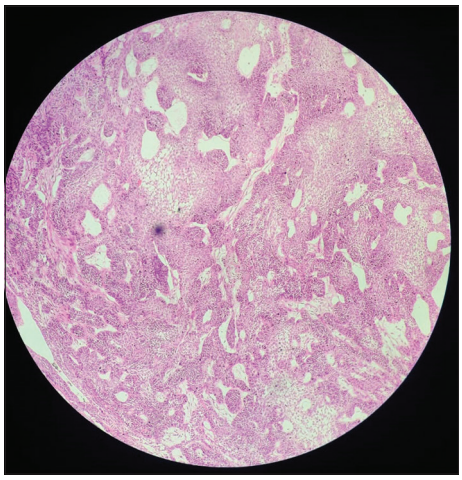

Figure 6: ×40 view showing the tall columnar cells with features of cytological atypia

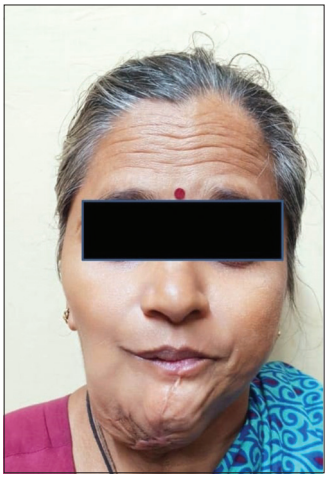

Figure 7: Post-operative view of patient

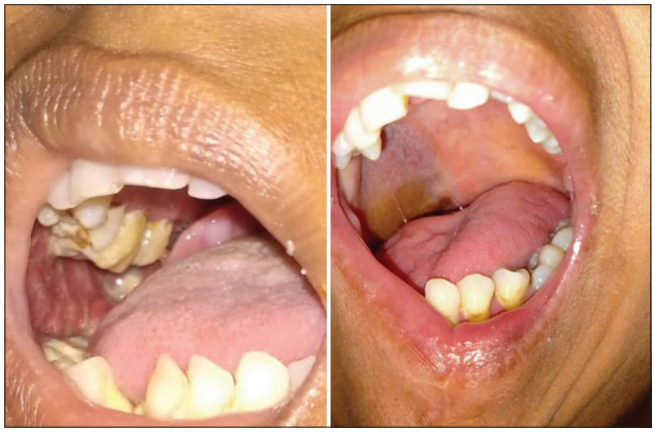

Figure 8: Pre-operative and post-operative mouth opening

The distinct histopathological features of ameloblastic carcinoma were nuclear enlargement with granular stippled nucleus, nuclear hyperchromatism, pleomorphism and increased mitotic activity with abnormal mitosis. Dyskeratosis and dystrophic calcifications were observed in some cases.[7,8] Different histopathological patterns such as highly differentiated squamous cell, basaloid and poorly poorly differentiated squamous cell carcinoma were noted. Seldom does ameloblastic carcinoma reveal clear cell differentiation. The histopathological report of the present case showed varying sized nests, trabeculae and islands of columnar epithelial cells with eosinophilic cytoplasm, round to oval hyperchromatic nuclei showing reverse polarisation.

Differential diagnoses of ameloblastic carcinoma are acanthomatous ameloblastoma, squamous odontogenic tumour, primary intraosseous carcinoma and clear cell odontogenic carcinoma.[5] The former has jigsaw puzzle type nesting of tumour cells, presence of stellate reticulum and distinctive cystic degeneration of nests.[6]

Two types of ameloblastoma can also be reviewed for the differential diagnosis. Acanthomatous ameloblastoma exhibits varying degrees of squamous metaplasia, keratinisation of the stellate reticulum of tumour islands. Keratoameloblastoma is a rare variant of ameloblastoma having prominent keratinising cysts.[8,9] Thus, the term ameloblastic carcinoma can be applied to our case, which showed focal histologic evidence of malignant disease including cytologic atypia and mitoses with indisputable features of classic ameloblastoma.

Surgical resection along with prophylactic excision of involved lymph nodes is the treatment of choice for ameloblastic carcinoma. *En bloc* removal with 1–2 cm of normal bone margin is the safest surgical modality. The local recurrence rates are <15% and recur 0.5–11 years after definitive therapy. Distant metastasis is usually fatal.[7,10]

Some authors suggested 2–3 cm bony margins removal in *en bloc* resection. Braimah *et al*., found the use of cisplatin, adriamycin and cyclophosphamide to be valuable.[6]

The patient presented here did not show any obvious clinical signs and symptoms in the follow-up period. To rule out distant metastasis, the patient underwent chest X-ray at the end of two years [Figure 7]. Mild improvement of mouth opening was noted [Figure 8].

Conclusion

Ameloblastic carcinoma is an aggressive neoplasm which is locally invasive although the rate of distant metastasis is rare. Early diagnosis and treatment give good prognosis.

Declaration of patient consent

The authors confirm that all appropriate consents are taken from the patient. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initial will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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