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CASE STUDY

Ameloblastic carcinoma of the mandible: A case report with a review of the literature

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Ameloblastic carcinoma is an uncommon and aggressive malignancy of odontogenic origin, distinguished by unique histological characteristics. It typically necessitates a more extensive surgical intervention compared to ameloblastoma. It predominantly affects the mandible and is observed across a broad age range. Accounting for approximately 1% of all jaw cysts and tumors, it is considered the most common and clinically significant odontogenic tumor. Selective regional lymph node dissection should be undertaken during neck dissection, and adequate time should be allowed before reconstructing the post-resection defect to address the possibility of tumor recurrence. In our case, we performed a supraomohyoid neck dissection for one such case reported to our department, followed by a successful reconstruction of the mandible with the reconstruction plate.

Keywords: malignant ameloblastoma, odontogenic tumor, squamous metaplasia, neck dissection, ameloblastic carcinoma, mandibular tumor, mandibular reconstruction

Introduction

Ameloblastoma is a benign tumor characterized by local invasiveness, commonly presenting as a painless, slow-growing swelling in the jaw. Around 80% of cases are found in the mandible. It may appear as a cystic lesion or a larger mass, sometimes associated with ulceration, extensive bone loss, and tooth mobility (1). Accounting for approximately 1% of all jaw cysts and tumors, it is considered the most prevalent and clinically important odontogenic tumor. While this neoplasm is primarily known for its local invasiveness and high recurrence rate, it rarely exhibits aggressive behavior or metastatic potential (2).

The potential for malignancy in ameloblastoma has been a topic of extensive discussion and debate over the years. While there is little disagreement that ameloblastoma exhibiting metastatic behavior is classified as malignant, it is noteworthy that such tumors often display a benign histologic appearance. Ameloblastomas are sometimes classified as malignant based solely on their aggressive clinical behavior, even without evidence of metastasis. These tumors often display atypical or uncommon histological features. Various terms, such as ameloblastic carcinoma, malignant ameloblastoma, primary intra-alveolar epidermoid carcinoma, and metastatic ameloblastoma, have been used to describe tumors originating from ameloblastomas. This article discusses an uncommon case of ameloblastic carcinoma of the lower jaw, specifically affecting the left ramus.

Case report

A 29-year-old male visited the department complaining of a persistent swelling in his left posterior mandible with intra-oral suppuration associated with four years. Before this, the patient had undergone extensive dental review along with the fine needle aspiration cytology (FNAC) of the same, but the patient is unaware of the report and has lost it.

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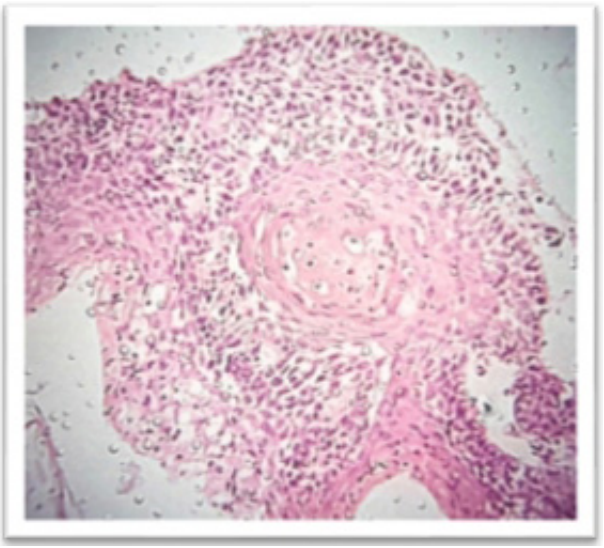

FIGURE 1 | Islands and sheets of epithelium.

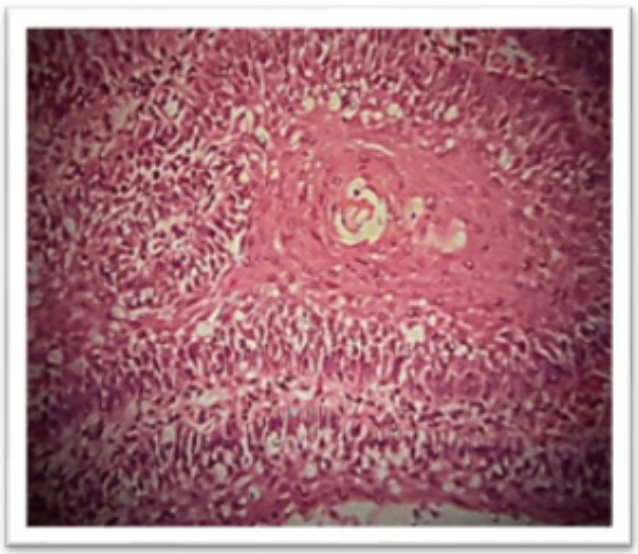

FIGURE 2 | Central cells resembling stellate reticulum within islands.

On clinical examination, extra-orally, a diffuse swelling was noticed on the left side face in the lower third region, extending anteroposteriorly along the body of the mandible up to the angle region and Superio-inferiorly from the left lobule of the ear up to the lower border of the mandible. Palpation revealed the swelling to be firm, diffuse, non-tender, non-fluctuant, non-reducible, non-pulsatile, non-compressible, afebrile surface temperature, and overlying skin appeared normal.

Regional lymph node examination revealed two submandibular lymph nodes being palpable, level Ib, having an anterior one sized to approximately 1 cm, mobile, soft, tender, and a posterior one sized to approximately 1 cm, fixed, firm, and non-tender.

Clinical examination intra-orally revealed vestibular obliteration present from the region of 37 to the retromolar area, having a whitish appearance with flattened borders, round ends, and margins that were non-indurated with palpatory findings to be firm, ill-defined, non-reducible, non-compressible, and non-pulsatile, with surface temperature afebrile.

Radiographic imaging of the jaws demonstrated a well-defined radiolucency involving the left side body and ramus of the mandible, with features of poorly defined borders and cortical plate breaching.

The histopathological examination showed epithelial islands and sheets arranged in an infiltrative pattern within a stroma of mature fibrous tissue, with a single layer of peripheral columnar cells observed. Few central cells resembling stellate reticulum that are condensed and hypercellular along with squamous metaplasia within the islands, nuclear enlargement, mild pleomorphism, hyperchromatism, increased mitotic activity, and altered nuclear-cytoplasmic ratio (**Figures 1** and **2**). The connective tissue exhibits mature collagen fibers accompanied by chronic inflammatory cells, regions of hemorrhage, and signs of degeneration, indicative of a malignancy of epithelial origin.

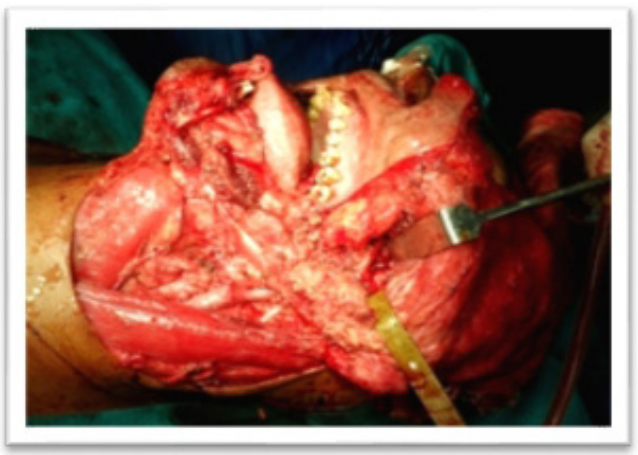

FIGURE 3 | Supraomohyoid neck dissection.

Under general anesthesia, the patient underwent wide local excision with supraomohyoid neck dissection on the left side, followed by primary closure (**Figures 3** and **4**). Additionally, mandibular reconstruction using a reconstruction plate was performed three months after resection to minimize the risk of recurrence (**Figures 5** and **6**). Postoperative oral physiotherapy, including jaw exercises and soft diet instructions, was introduced to restore function and prevent fibrosis of the reconstructed mandible.

Discussion

The differentiation in epithelial neoplasms is often regarded as a key factor in predicting metastatic behavior. A primary

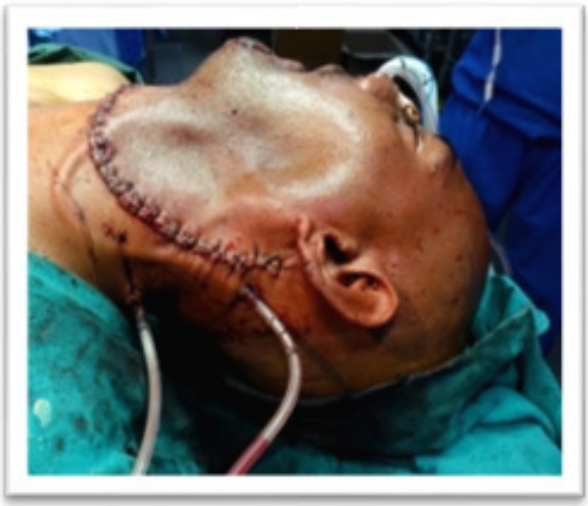

FIGURE 4 | Closure with drain placed after surgery.

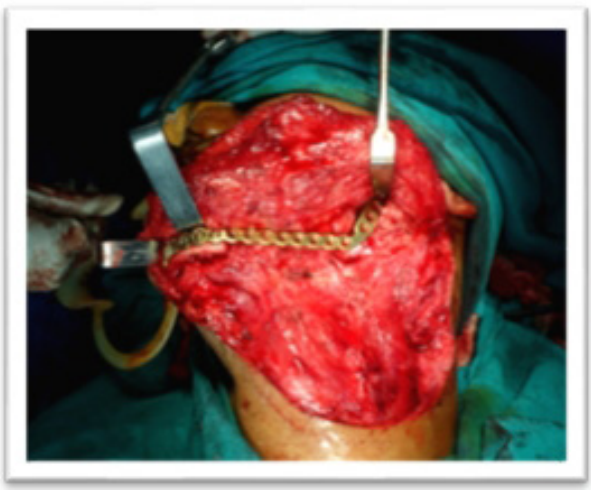

FIGURE 5 | Reconstruction plate fixed.

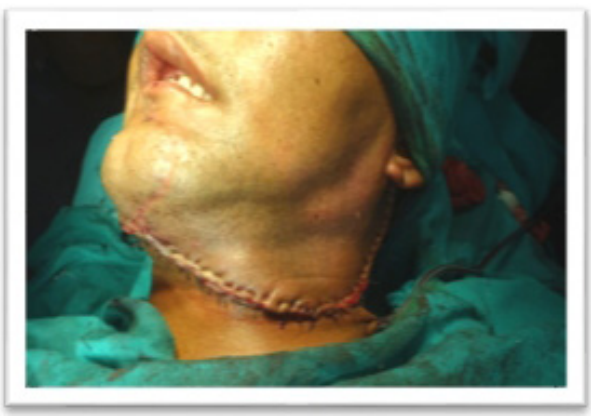

FIGURE 6 | Closure done after reconstruction.

distinction between the classification systems of Elzay and Slootweg and Müller lies in their subtle emphasis on histogenesis (3). These authors suggest that the term "ameloblastic carcinoma" should be applied to lesions exhibiting histological features characteristic of both ameloblastoma and carcinoma. Such tumors may metastasize, with malignant histological traits appearing in the primary lesion, the metastatic deposits, or both (4). Ameloblastic carcinoma is reported to occur at twice the rate of malignant ameloblastoma, with a prevalence ratio of 2:1 (3).

To describe a tumor from the malignant transformation of epithelial cells, initially, Shafer et al. introduced the term "ameloblastic carcinoma" (5). Over time, the differentiation between malignant ameloblastoma and ameloblastic carcinoma has been a topic of significant debate and has undergone various classifications.

The differentiation between these two entities is based on their histological and metastatic characteristics. Malignant ameloblastomas exhibit well-differentiated, benign histological features in primary and metastatic sites, whereas ameloblastic carcinomas are characterized by cytological atypia.

In 1982, Elzay highlighted that the WHO classification did not clearly differentiate between histologically similar tumors to classic ameloblastoma and lesions that metastasize from ameloblastoma-like tumors with malignant histological features before metastasis. He recommended revising the classification system, proposing that all primary intraosseous carcinomas unrelated to the salivary glands be classified as primary intraosseous carcinomas (PIOCs).

PIOC describes a carcinoma of the jaw that lacks the histological characteristics of ameloblastoma and does not develop from an odontogenic cyst. The category "other carcinomas" encompasses those arising from odontogenic epithelium, including cases associated with odontogenic cysts (6).

Recent studies have documented similar cases of ameloblastic carcinoma requiring aggressive surgical intervention. Brukas et al. (7) emphasized the necessity of extensive excision and delayed reconstruction to mitigate recurrence risks. Another report by Schliephake et al. (8) highlighted successful long-term outcomes following mandibular reconstruction with vascularized grafts after a waiting period of 3–6 months. These studies reinforce the importance of our treatment approach, emphasizing staged reconstruction and diligent follow-up.

A definitive histological standard for diagnosing ameloblastic carcinoma remains uncertain. Hall et al. established distinct criteria for its identification. The findings emphasize the presence of epithelial islands, sheets, or trabeculae, with absent stellate reticulum areas. Additional malignant features observed include hyperchromatic nuclei, prominent or atypical nucleoli, increased mitotic activity, necrosis, calcification, and neural and vascular invasion evidence (9).

In the differential diagnosis of ameloblastic carcinoma, two types of classic ameloblastoma should also be considered.

Acanthomatous ameloblastoma is characterized by differing degrees of squamous metaplasia and keratinization occurring in the stellate reticulum region of the tumor islands. Despite this, it maintains peripheral palisading and lacks cytological characteristics indicative of malignancy. Kerato-ameloblastoma, on the other hand, is an uncommon variant of ameloblastoma characterized by extensive keratinizing cysts. This atypical presentation can create diagnostic challenges, potentially preventing the pathologist from recognizing its otherwise ameloblastomatous nature (10).

Ameloblastic carcinoma, in all its forms, follows an aggressive progression characterized by significant local destruction and a risk of metastasis. While a standardized treatment protocol is lacking, wide local excision with adequate hard and soft tissue margins is generally recommended. In this instance, a simultaneous neck dissection was performed for both diagnostic staging and therapeutic purposes. Adjuvant radiotherapy can be beneficial in situations involving close or positive margins or nodal metastases, although chemotherapy has shown no clear benefit. Given the tendency of these tumors to recur, close and long-term follow-up is essential, including regular evaluations and imaging, particularly of the chest.

Conclusion

This case demonstrates malignancy of epithelial origin, characterized by stellate reticulum and squamous metaplasia within the tumor islands. The patient underwent successful radical surgical resection followed by reconstruction of the defect using a reconstruction plate. The 7-month follow-up, patient remains disease-free, including 2 months' post-reconstruction. Ameloblastoma exhibits a broad spectrum of histological and biological behavior, ranging from benign forms to overt malignancy. Post-resection, the defect reconstruction can proceed as expected after any carcinoma resection. However, an adequate waiting period should be observed before reconstruction due to the potential for tumor recurrence.

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