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Case Report: Metastatic Ameloblastic Carcinoma with Pulmonary, Rib, and Pericardial Metastases and Subsequent Management of Rib Pain

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ABSTRACT

Ameloblastoma, a benign yet locally aggressive odontogenic tumor, can rarely undergo malignant transformation to ameloblastic carcinoma (MAC), a highly aggressive malignancy with a propensity for distant metastasis. While lungs are common metastatic sites, involvement of the pericardium is exceedingly rare. This case report details the challenging clinical course of a 71-year-old male veteran who initially presented with maxillary ameloblastoma in 2019. Despite initial treatment, he experienced recurrence in 2024, which histologically demonstrated transformation to ameloblastic carcinoma with extensive invasion. Subsequently, he developed symptomatic metastatic disease, including a biopsy-proven lesion in the left fourth rib requiring palliative radiation, discrete pulmonary nodules, and a 1.9 cm pericardial metastasis that also received external beam radiation. The patient's complex journey highlights diagnostic delays, the aggressive nature of MAC, the critical need for a multidisciplinary approach, and systemic barriers in accessing timely specialized care. This case underscores the importance of vigilant long-term surveillance for ameloblastoma and aggressive management strategies for its malignant transformation, especially when rare metastatic patterns emerge.

Keywords: ameloblastic carcinoma, odontogenic tumors, bone metastasis, pericardial metastasis, palliative radiation therapy

INTRODUCTION

Ameloblastoma, a benign but locally aggressive odontogenic tumor, typically arises in the mandible or maxilla and is known for its high recurrence rate if not adequately resected [1]. While generally considered benign, a variant termed metastasizing ameloblastoma (METAM) demonstrates distant metastasis despite benign histological features [1]. Malignant transformation to ameloblastic carcinoma, as seen in this case, is exceedingly rare but confers a significantly worse prognosis due to its potential for local aggression and distant metastasis. Metastatic ameloblastic carcinoma (MAC) commonly spreads to the lungs and regional lymph nodes, with bone involvement being less frequent [1]. The

occurrence of pericardial metastasis is rare and indicates advanced disease [1]. We present a challenging case of a 71-year-old male army veteran with recurrent ameloblastoma that transformed into ameloblastic carcinoma, exhibiting pulmonary, rib (necessitating palliative radiation for pain), and pericardial metastases, highlighting the complex management required for this aggressive entity.

CASE PRESENTATION

A 71-year-old White male army veteran from the Orlando VA Health System, initially presented in November 2019 with an ameloblastoma of the right maxilla. He underwent a right maxillectomy with reconstruction of the right zygoma and right inferior orbital floor, including implant hardware, and a right neck dissection. Following this surgery, he was lost to follow-up by the VA.

He remained asymptomatic until 2023, when he developed increasing right maxillary sinus pressure. This prompted a diagnostic workup at the VA, including an MRI of the face on February 5, 2024, which revealed significant postsurgical changes in the right maxillary area. The MRI showed complete opacification of the right maxillary sinus and nasal cavity, with associated erosion of the right maxillary sinus wall and extension into the premaxillary and retroantral spaces, as well as the right pterygopalatine fissure.

On May 21, 2024, the patient underwent a second right maxillary resection. Pathology identified a recurrent adenoid ameloblastoma with extensive invasion and osteomyelitis. During this procedure, the hardware prosthesis was explanted. Despite pathological confirmation of non-malignant stratification at this stage, postoperative radiation treatment was recommended due to the tumor's extensive invasion and unresectability. A September 24, 2024 4-month follow-up CT and MRI of the face confirmed a status of No Evidence of Disease (NED).

However, surveillance scans on March 20, 2025, at another medical facility, showed recurrent soft tissue disease in the right maxillary region with associated inflammatory changes on MRI. A repeat MRI on April 8, 2025, confirmed the soft tissue density in the right maxillary sinus, now slightly larger than the previous study, with apparent extension into the right anterior nasal cavity. Associated CT images corroborated these findings.

On April 15, 2025, the patient underwent surgical resection at a specialized cancer center. Resection of the right nasal sinus content confirmed recurrent adenoid ameloblastoma with carcinomatous transformation, now diagnosed as ameloblastic carcinoma. Histopathological examination revealed areas of necrosis and an elevated mitotic rate of up to 20 mitoses per 10 high-power fields (HPF), consistent with malignant transformation. Following this surgery, he received adjuvant radiation therapy to the right maxilla, completing 68.4 Gray on July 15, 2025.

Subsequent to this, the patient developed rib pain, which prompted further investigation. This led to the diagnosis of a biopsy-proven metastatic lesion in the left fourth rib, for which he received stereotactic body radiation therapy (SBRT) to 30 Gray, concluding on August 13, 2025. This radiation was specifically administered for palliation of pain associated with the bone metastasis.

A PET/CT scan performed on August 7, 2025, revealed several metastatic foci:

A pleural-based mass in the left lateral chest wall at the fourth rib, measuring 4.1 cm with an SUV of 5.1.

A discrete left lower lobe lateral lesion, 2.4 cm with an SUV of 6.4.

A posterior mediastinal lesion in the left periaortic region adjacent to the pericardium, measuring 1.9 cm with an SUV of 6.4.

A small nodule in the right lower lobe posteriorly at the lung base, measuring 6 mm with an SUV of 2.4.

A 3.5 cm soft tissue density in the right maxilla with an SUV of 4.3, indicating persistent or recurrent disease at the primary site.

As of September 18, 2025, the patient was scheduled for a surgical procedure on October 7, 2025, at the VA Hospital for reconstruction of the right lower eyelid and to fill the zygomatic arch defect, to be performed by plastic surgery.

During a follow-up on October 16, 2025, the patient reported anterior chest pain, likely related to the pericardial mass, but otherwise felt well. He expressed continued interest in plastic and reconstructive surgical options for the right maxilla defect. While a referral to medical oncology had been made, he had not yet seen them.

On December 11, 2025, the patient completed external beam radiation therapy to the left pericardium mass. He reported feeling generally well, with no specific complaints of chest pain, discomfort, or shortness of breath. He noted ongoing depression, for which his current escitalopram 20 mg daily seemed refractory. He was also actively seeking an oculofacial plastic surgeon for reconstruction of his right lower eyelid and was awaiting VA approval for his medical oncology referral, highlighting systemic delays in comprehensive care.

Figure 1A

Figure 1B

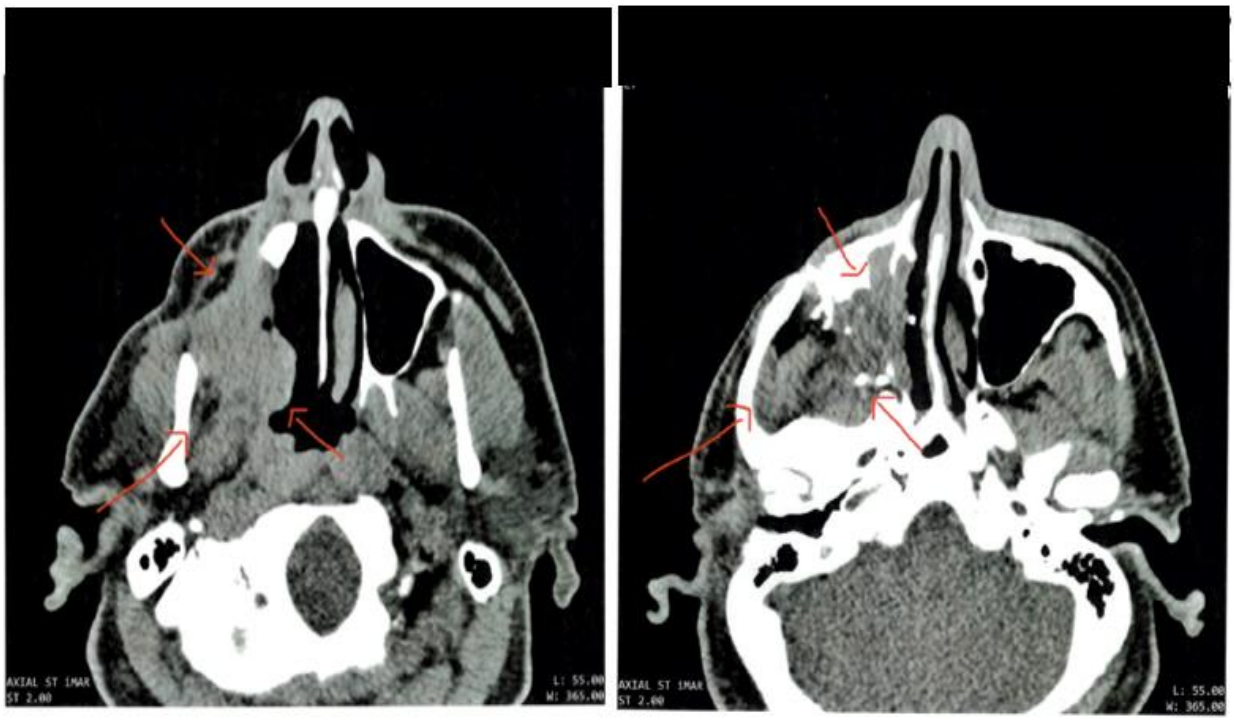

Figure 1A and 1B: Soft tissue mass in right maxillary sinus with complete obstruction of right frontal and right ethmoid sinus.

DISCUSSION

This case illustrates the aggressive nature and challenging clinical course of ameloblastic carcinoma, particularly when it transforms from a recurrent ameloblastoma. While conventional ameloblastomas are locally destructive, their malignant counterparts, though rare, have a high propensity for distant metastasis, most commonly to the lungs [1-3]. The patient's case demonstrates this metastatic potential, with several discrete pulmonary nodules, rib lesion, and a small pericardial lesion, indicative of systemic spread. According to a recent clinicopathological review by Pandiar et al., lungs are the most commonly affected secondary sites, found in 29 out of 42 cases, often bilaterally, while pericardial involvement is rare, reported in only one case within their extensive review [1]. Bone metastases, such as the patient's rib lesion, are also infrequent but documented [1]. The overall incidence of malignant ameloblastoma (which can include both METAM and ameloblastic carcinoma) has been reported at 1.79 per million/person.

The initial misinterpretation of the recurrent lesion as "non-malignant stratification" despite its extensive invasion, followed by later confirmation of carcinomatous transformation, highlights the diagnostic challenges and the critical importance of repeated biopsies and multidisciplinary review in the context of persistent or growing lesions. The 2017 WHO classification of head and neck tumors re-classified METAM under benign epithelial odontogenic tumors, although it retains a malignant ICD-O code, while ameloblastic carcinoma is distinctly characterized by malignant features such as increased nucleo-cytoplasmic ratio, pleomorphism, nuclear hyperchromatism, and increased mitotic activity [1,4]. The patient's case is classified as ameloblastic carcinoma due to the "carcinomatous transformation" with malignant features observed in the maxillary recurrence, indicating a more aggressive biology than typical METAM. The development of severe rib pain, which proved to be a metastatic focus requiring palliative radiation, further emphasized the systemic nature of the disease and the importance of comprehensive screening for metastases in such cases.

The management of metastatic ameloblastic carcinoma is complex, often involving combinations of surgery, radiation, and systemic therapies. However, systemic treatment options for MAC are limited and often less effective compared to local control measures. In the patient's case, the primary strategy has been local control through repeated surgeries and targeted radiation to metastatic sites (axilla, rib, pericardium). The need for palliative radiation to the rib for pain management underscores the importance of addressing patient symptoms and improving quality of life in the context of advanced disease. Pandiar et al.'s review highlights that inadequate treatment of the primary lesion can result in multiple recurrences and, in rare instances, metastasis [1]. The mean interval between the primary and the secondary tumor in their review was 11.45 years (range 0-35 years), suggesting that a long latent period can occur, which is consistent with the patient's timeline from initial diagnosis to widespread metastasis [1-3]. Previous reviews have also noted varying mean ages of occurrence, with more recent data suggesting a trend towards older patients, which aligns with the patient's presentation [1-3].

The recurrent delays in obtaining consistent follow-up, both for oncological assessment and reconstructive surgery, significantly complicated the patient's care. The prolonged wait for a medical oncology referral, despite clear evidence of metastatic disease, points to systemic barriers within healthcare delivery that can hinder timely, integrated management for patients with rare and aggressive cancers. These delays not only impact the potential for effective

treatment but also contribute significantly to patient distress. Furthermore, the patient's reported refractory depression highlights the profound psychological burden associated with an advanced, recurrent, and metastatic cancer diagnosis, emphasizing the critical need for integrated supportive and palliative care throughout the disease trajectory.

CONCLUSION

This case of the patient presents a rare instance of recurrent ameloblastoma transforming into metastatic ameloblastic carcinoma with focal pulmonary, symptomatic rib (requiring palliative radiation), and a discrete pericardial metastasis. It underscores several critical learning points: the importance of diligent long-term follow-up for ameloblastoma, the necessity of considering malignant transformation in recurrent or aggressive lesions, and the critical role of a multidisciplinary team for timely diagnosis and comprehensive management. The presence of these metastases, including the uncommon pericardial involvement, signifies a challenging disease course, aligning with the understanding that even benign-appearing ameloblastomas can metastasize (METAM), and their malignant counterparts (ameloblastic carcinoma) are particularly aggressive [1]. Delays in care, whether diagnostic, therapeutic, or reconstructive, compounded by challenges in access to specialized care, can significantly impact patient outcomes and quality of life in such aggressive malignancies. This case advocates for improved coordination of care and immediate access to specialized oncology services, including robust supportive and palliative care, for patients with rare and complex cancers.

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