

Multimodal treatment for multifocal metastatic maxillary adenoid cystic carcinoma: A case report

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Abstract. Maxillary adenoid cystic carcinoma (ACC) is rare, with pleural and brain metastases representing an unprecedented presentation. The present case report illustrates the challenges and potential for prolonged survival in such an advanced stage of malignant disease. A 52-year-old male patient presented with a left maxillary mass, initially misdiagnosed as ameloblastoma; subsequently, it was confirmed as maxillary ACC (T4aN0M0, stage IVA). Following primary tumor resection and radiotherapy, the patient developed pleural (February 2021) and brain metastases (March 2022). A multimodal treatment strategy was used, involving aggressive surgical resection of the primary tumor, comprehensive radiotherapy, systemic chemotherapy regimens and adjuvant targeted molecular therapy. By using this sequential multimodal therapy, the patient achieved long-term survival exceeding 5 years. The present case report highlights the potential for favorable, prolonged survival even in the presence of multifocal metastatic maxillary ACC when managed with a persistent, sequential and tailored combination of modern therapeutic modalities.

Introduction

Adenoid cystic carcinoma (ACC) is a malignant tumor that originates from the secretory glands, accounting for only 1% of head and neck malignancies. It most commonly arises in the salivary glands, where it constitutes ~30% of all salivary gland malignancies worldwide (1). Known risk factors remain to be elucidated, although prior radiation exposure and certain

genetic alterations (such as MYB-nuclear factor I B) fusion and NOTCH1 mutations) have been associated with ACC (2,3). ACC exhibits distinct biological behaviors: Slow growth but high propensity for perineural invasion, local recurrence and distant metastasis (4). The primary treatment for localized ACC is surgical resection with postoperative radiotherapy (4). However, metastatic ACC has a poor prognosis due to limited systemic options. Palliative chemotherapy regimens (such as cisplatin-based or anthracycline-containing combinations) and molecular targeted agents (lenvatinib) are commonly used; however, objective response rates remain modest, typically ranging between 10 and 30%, underscoring the need for more effective therapeutic strategies (5,6).

In the present study, a case report of maxillary ACC with distant metastases to the pleura and brain is reported. The patient underwent multimodal treatment, including surgery, radiotherapy, chemotherapy and targeted therapy.

Case report

This case report was prepared in accordance with the CARE guidelines for case reports (7). A 52-year-old male farmer of Han Chinese ethnicity presented to Nanchong Central Hospital (Nanchong, China) in January 2020 with a painless, slowly enlarging left maxillary mass. Family history was negative for malignancies. As the primary wage-earner for their household, the main concern of the patient was whether functional impairment would affect their work capacity.

Physical examination demonstrated a lack of mucosal lesions, such as ulcers, on the palate, buccal mucosa or gingiva. No cervical lymphadenopathy was noted. A panoramic dental X-ray (Fig. 1A) and maxillofacial CT scan (Fig. 1B) were performed, and initially left maxillary ameloblastoma was suggested. The panoramic radiograph (Fig. 1A) indicated expansile osteolysis indistinguishable from ameloblastoma. The lesion primarily involved the left maxillary alveolar process and body, forming a soft tissue mass measuring ~4.7x4.2 cm with expansile growth on CT imaging (Fig. 1B). The patient underwent total resection of the left maxilla, partial resection of the right maxilla, implantation of a bioabsorbable membrane, formation of a fascial flap and exploration of the deep cervicofacial mass. Intraoperatively, the tumor was demonstrated to have invaded the anterior and posterior lateral walls of the maxillary sinus, completely filling the sinus.

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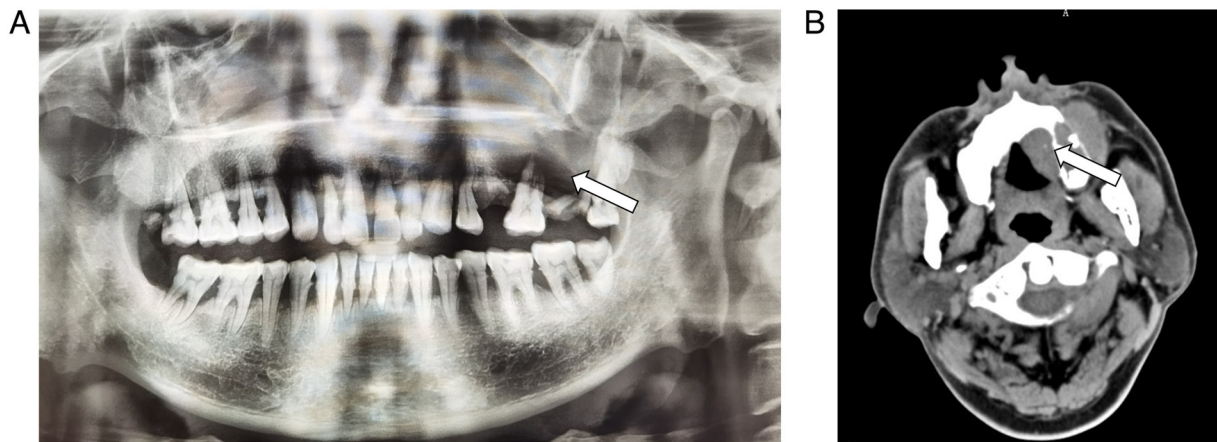


Figure 1. Initial imaging findings of the left maxillary mass. (A) Oral panoramic radiograph indicating an oval osteolytic lesion ~3 cm in size. (B) Maxillofacial CT indicates a localized osteolytic lesion in the left maxilla involving adjacent alveolar bone and left maxillary sinus floor, with a centered 4.7x4.2 cm soft tissue mass.

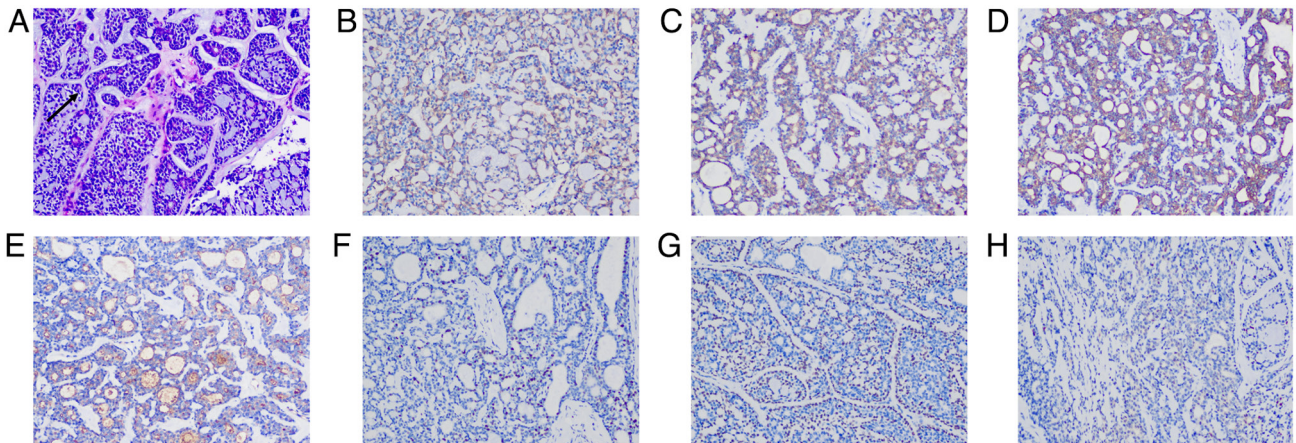


Figure 2. Histopathological and immunohistochemical features of primary adenoid cystic carcinoma. (A) Tumor cells are arranged in nests of varying sizes, exhibiting a cribriform pattern with cystic spaces containing basophilic mucoid material, reminiscent of a lotus root cross-section. A layer of mucin-secreting myoepithelial cells surrounds the cystic spaces (magnification, x100). (B) Positive for actin (magnification, x100). (C) Positive for CD117 (magnification, x100). (D) Positive for cytokeratin 8/18 (magnification, x100). (E) Positive for epithelial membrane antigen. (F) Ki-67 proliferation index is 20% (magnification, x100). (G) Positive for P63 (magnification, x100). (H) Positive for S-100 (magnification, x100).

Postoperative pathological diagnosis confirmed left maxillary ACC with negative margins and perineural invasion. Tumor cells were arranged in nests of varying sizes and exhibited a cribriform pattern with cystic spaces containing basophilic mucoid material. A layer of mucin-secreting myoepithelial cells surrounded the cystic spaces (Fig. 2A). Immunohistochemical analysis demonstrated the following results: CD117(+), epithelial membrane antigen (EMA)(+), cytokeratin (CK)8/18(+), P63(+), actin(+), S-100(+), Ki-67 (20% +) (Fig. 2B-H).

Tissue samples were fixed in 4% neutral buffered formalin at room temperature for 24-48 h, embedded in paraffin, and sectioned at a thickness of 4 μ m. Hematoxylin and eosin staining was performed at room temperature: sections were stained with hematoxylin for 5-7 min, followed by eosin for 1-2 min.

For immunohistochemical analysis, 4- μ m-thick paraffin-embedded sections were deparaffinized in xylene and rehydrated through a graded ethanol series. Heat-induced antigen retrieval was performed using citrate buffer (pH 6.0)

at 95-100°C for 20 min. Endogenous peroxidase activity was quenched by incubation with 3% hydrogen peroxide for 15 min at room temperature. Non-specific binding was blocked with 5% normal mouse/rabbit serum (Fuzhou Maixin Biotech, China) for 30 min at room temperature. Sections were then incubated with primary antibodies (1:100-1:200) overnight at 4°C. The primary antibodies used were as follows: CD117 (cat# Kit-0029), EMA (clone: E29; cat# Kit-0011), CK8/18 (clone: MX004+MX035; cat# MAB-1002), P63 (clone: MX013; cat# MAB-0694), actin (clone: MX083; cat# MAB-0871), S-100 (clone: MXR034; cat# RMA-1075), Ki-67 (clone: MXR002; cat# RMA-0731), CK (clone: MX005; cat# MAB-0671), vimentin (clone: MX034; cat# MAB-0735), calponin (clone: MX023; cat# MAB-0712), TTF-1 (clone: MX011; cat# MAB-0599), and PD-L1 (clone: MXR006; cat# RMA-0894); all from Fuzhou Maixin Biotech, China. After washing, sections were incubated with an HRP-conjugated anti-mouse/rabbit IgG secondary antibody (ready-to-use; cat# KIT-5005, Fuzhou Maixin Biotech, China) for 1 h at room

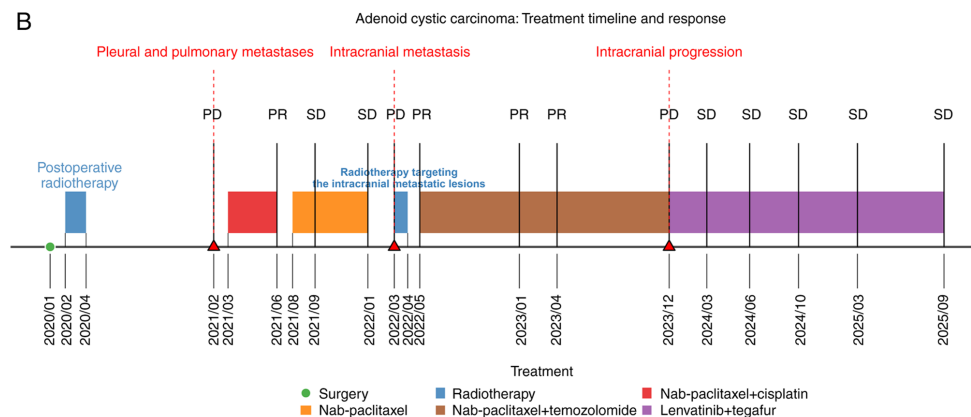
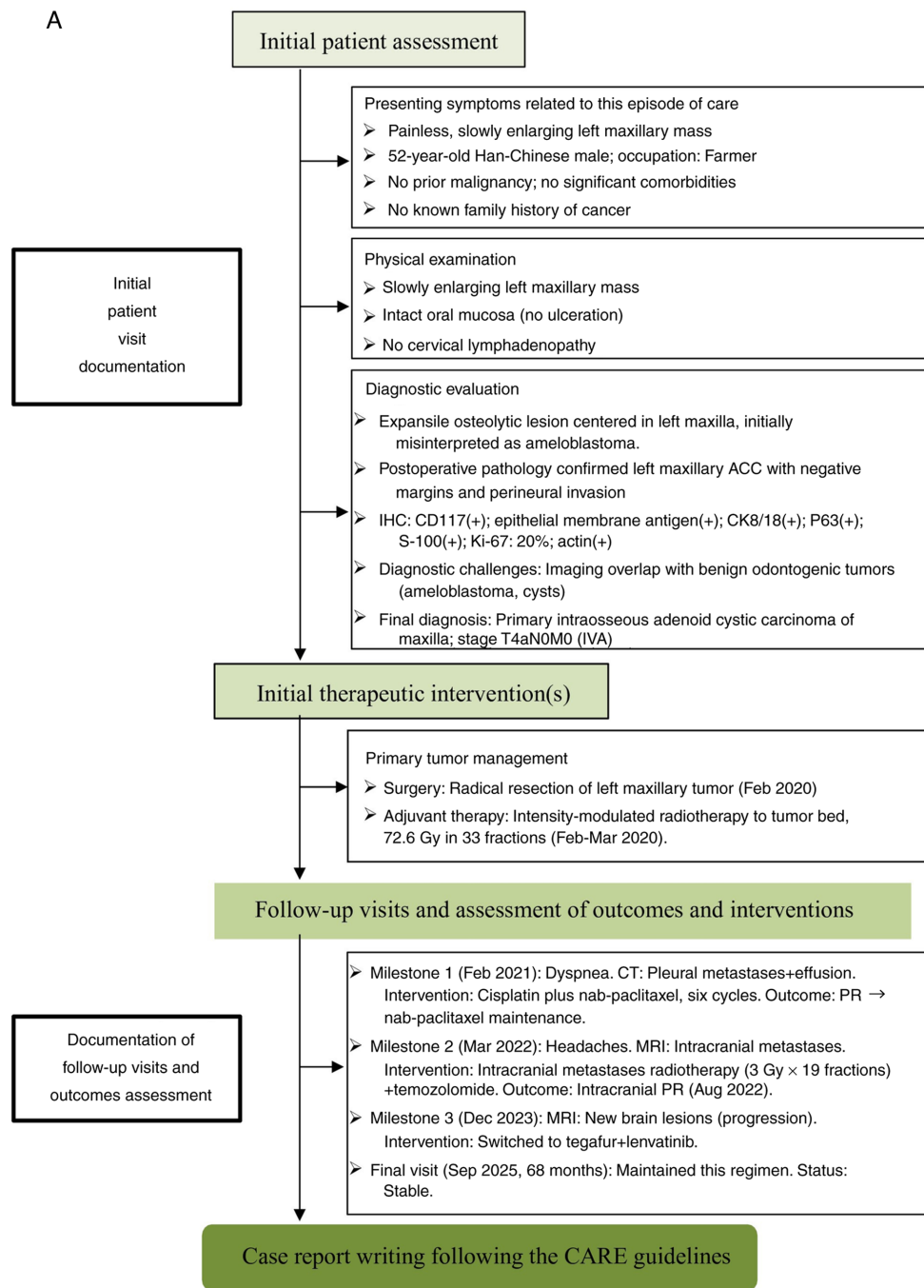


Figure 3. Treatment strategy and clinical course. (A) Outline of a CARE-guided workflow for comprehensive case reporting, encompassing initial assessment, therapeutic interventions and longitudinal follow-up of this rare malignancy. (B) Timeline illustrating the sequential adaptation of multimodal therapy for pleural and intracranial metastases, achieving sustained disease control and 68-month overall survival. ACC, adenoid cystic carcinoma; IHC, immunohistochemistry; CK, cytokeratin; MRI, magnetic resonance imaging; PR, partial response; PD, progressive disease; SD, stable disease.

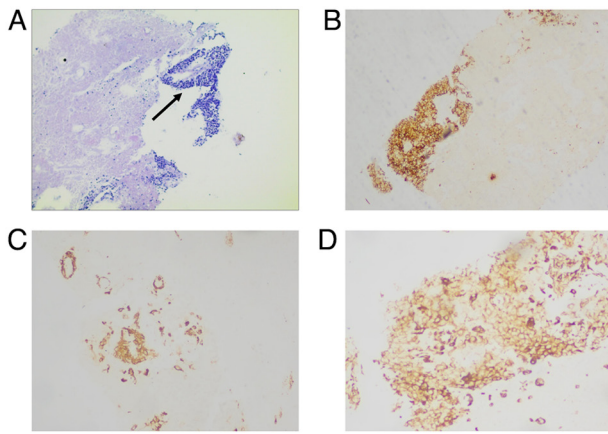


Figure 4. Histopathological confirmation of pleural metastasis. (A) Poorly differentiated tumor cell components demonstrating significant atypia against a background of necrosis (magnification, x100). (B) Positive for cytokeratin 8/18 (magnification, x100). (C) Weakly positive for smooth muscle actin (magnification, x100). (D) Positive for vimentin (magnification, x100).

temperature. Signal detection was performed using DAB (cat# TT-0805, Fuzhou Maixin Biotech, China). Sections were counterstained with hematoxylin for 1-2 min at room temperature. All slides were examined under a light microscope (Olympus BX43; Olympus Corporation, Japan).

Based on the postoperative pathology, the patient was diagnosed with ACC, stage T4aN0M0 (IVA) according to the 8th edition of the AJCC Cancer Staging Manual (8), and underwent adjuvant radiotherapy at the General Hospital of Western Theater Command of the Chinese People's Liberation Army (Chengdu, China) in February 2020. Radiotherapy was delivered as intensity-modulated radiation at 72.6 Gy in 33 fractions. Grade 1 mucositis, graded according to the Common Terminology Criteria for Adverse Events (CTCAE) version 5.0 (9), occurred but was resolved with supportive care. Fig. 3A and B present the entire treatment process of the patient as well as the timeline.

In February 2021, the patient developed dyspnea and orthopnea. A chest CT scan at an external hospital revealed multiple enhancing nodules on the right pleura with massive pleural effusion. The patient underwent closed thoracostomy drainage at the General Hospital of Western Theater Command, yielding hemorrhagic pleural fluid. Pleural biopsy pathology was suggestive of metastatic carcinoma (Fig. 4). The immunohistochemical results of the biopsy specimen were as follows: Carcinoma cells positive for CK, Ki-67 (10%+), smooth muscle actin and vimentin (Fig. 4); and negative for calponin, P63, S-100, thyroid transcription factor 1 (TTF-1) and programmed death-ligand 1 (Fig. S1). Due to the extreme fading and the limited quantity of tissue obtained from the pleural biopsy, immunohistochemical staining images for Ki-67 and P63 are not presented.

Although the pleural biopsy showed poorly differentiated carcinoma (Fig. 4A), differing from the cribriform pattern of the primary ACC (Fig. 2A), both lesions exhibited an infiltrative growth pattern with focal stromal collagenization. The immunohistochemical discrepancy (P63/S-100 negative in pleura vs. positive in primary ACC) likely reflects tumor dedifferentiation at the metastatic site. Alternative diagnoses,

including primary pleural mesothelioma and metastatic lung adenocarcinoma, were considered but excluded: Mesothelioma was ruled out by negative calponin and absence of mesothelial morphology, and lung adenocarcinoma was unlikely due to TTF-1 negativity. The chronological progression from primary ACC to pleural metastasis, coupled with the absence of other primary tumors, further supports the diagnosis of metastatic ACC.

Due to the pleural metastasis, every 21 days the patient received six cycles of cisplatin (75 mg/m² IV, days 1-3) combined with nab-paclitaxel (260 mg/m² IV, day 1) regimen chemotherapy, achieving partial response. Grade 2 neutropenia (according to CTCAE version 5.0) occurred and was managed with granulocyte colony-stimulating factor (G-CSF; 5 µg/kg subcutaneously for 3-5 days until neutrophil recovery). Grade 1 peripheral neuropathy resolved post-treatment. Maintenance nab-paclitaxel (260 mg/m² every 21 days) continued for 8 months until intracranial progression. The thoracic lesion remained stable (Fig. 5).

In March 2022, the patient underwent cranial magnetic resonance imaging (MRI) for headaches, revealing possible brain metastases. Radiotherapy targeting the intracranial metastatic lesions commenced within one week following the MRI (57 Gy in 19 fractions). Considering the poor central nervous system penetration of previous chemotherapeutic agents, temozolomide (75 mg/m² orally, once daily) was added to the nab-paclitaxel regimen and continued until disease progression. No grade ≥3 adverse events (according to CTCAE version 5.0) occurred; only grade 1 nausea was reported. Follow-up imaging in August 2022 indicated partial response of the intracranial lesions (Fig. 6).

The patient continued regular surveillance. In December 2023, follow-up cranial MRI revealed new intracranial lesions, indicating resistance to second-line chemotherapy and intracranial progression. Due to decreased treatment compliance associated with prolonged disease, therapy was switched to tegafur (40 mg/m² orally twice daily, days 1-14) combined with lenvatinib (8 mg orally daily), an anti-angiogenic targeted agent. Disease evaluations in October 2024 and March 2025 confirmed stable disease (Fig. 7).

The patient maintained this regimen with ongoing surveillance. The last follow-up was conducted in September 2025, at which time the patient was alive. In terms of patient perspective, the 5-year survival milestone allowed the patient to witness their daughter's high school graduation and university enrollment moments. Overall, the treatment provided to the patient highlighted that advanced cancer is not necessarily incurable when innovative therapies are persistently pursued.

Discussion

ACC is a rare, slow-growing and often asymptomatic malignancy. Intraosseous ACC (IACC) accounts for only <0.4% of all ACCs, with cases in the maxilla being even rarer (10). The pathogenesis of IACC remains to be elucidated; however, ACC typically arises from the secretory glands, suggesting that primary intraosseous ACC may originate from ectopic salivary tissues during mandibular embryogenesis (11). Although typically indolent, ACC exhibits perineural invasion, allowing the spread along nerves, leading to recurrence and metastasis. Hematogenous

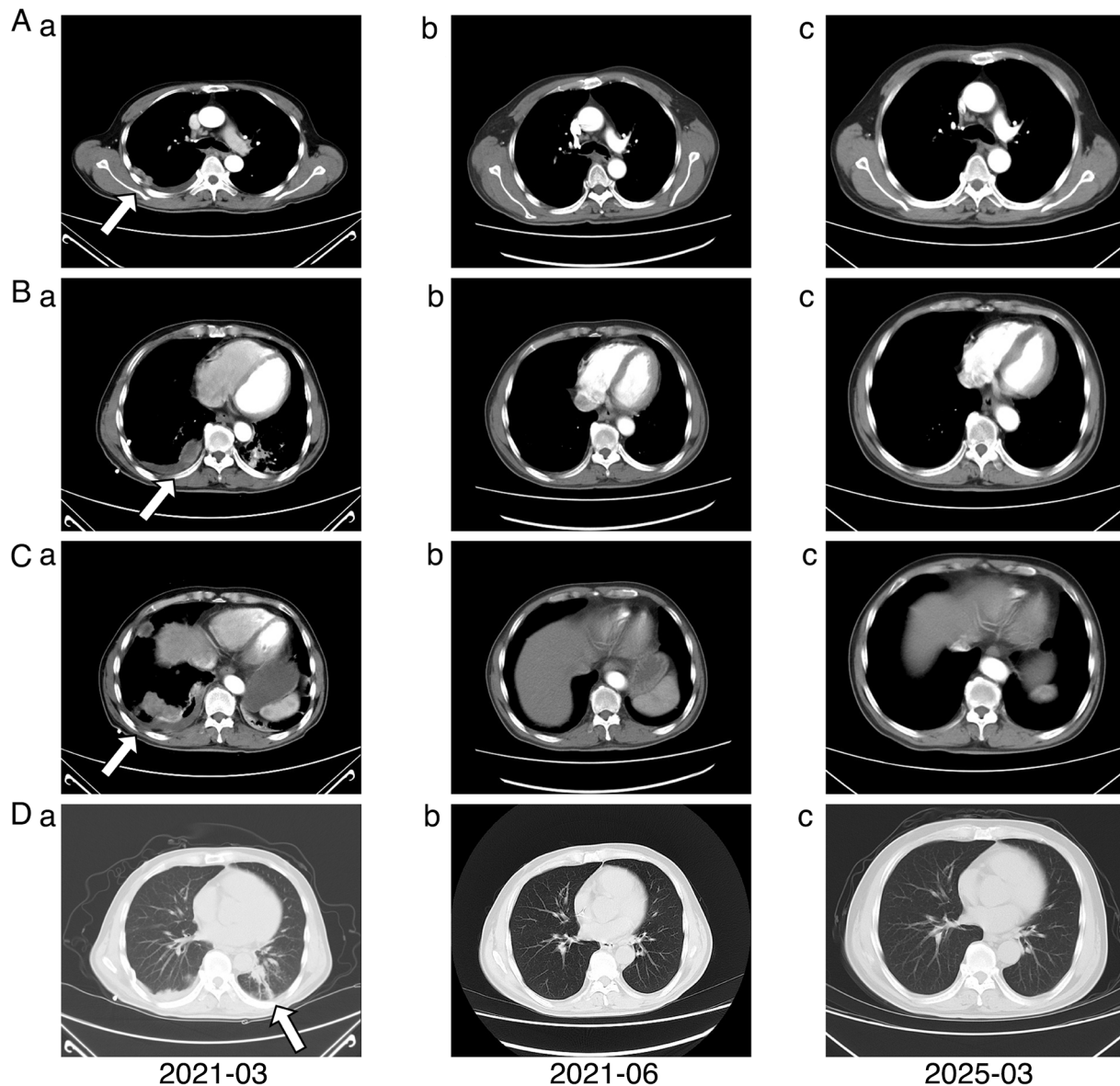


Figure 5. Serial chest CT demonstrating treatment response of pleural metastases. March 2021: (Aa-Ca) Multiple irregular, enhancing pleural masses in the right chest wall (arrows); (Da) a soft tissue nodule in the left lower lobe posterior basal segment invading adjacent bronchus, with distal obstructive inflammation. June 2021: (Ab-Db) Notable reduction in right pleural and left lung lesions. (Ac-Dc) March 2025: No new lesions in right pleura or lungs.

spread to the lungs and bones is most common (12), while pleural and brain metastases are rare and have not, to the best of our knowledge, been previously reported in maxillary ACC.

The observed immunohistochemical discordance, specifically the loss of myoepithelial markers P63 and S-100 in the metastatic pleural lesion, is a particularly noteworthy finding. This phenotypic alteration aligns with high-grade transformation or dedifferentiation, a recognized pattern of tumor progression in advanced ACC that often leads to loss of lineage-specific markers at metastatic sites (13,14). Such alterations reinforce the necessity of integrating clinical context and histomorphology with biomarker interpretation to avoid diagnostic ambiguity.

Furthermore, the non-specific clinical manifestations of ACC, such as a slow-growing mass accompanied by pain and swelling, often mimic odontogenic lesions (such as odontogenic cysts, ameloblastoma and apical periodontitis), leading to misdiagnosis. The erroneous initial diagnosis of ameloblastoma

underscores the diagnostic complexity of maxillary ACC. In the present case, panoramic radiography demonstrated expansile osteolytic changes with cortical thinning, which are features indistinguishable from ameloblastoma. This diagnostic pitfall reinforces that histopathological verification remains mandatory for expansile maxillary lesions, regardless of benign-appearing imaging characteristics (15). Therefore, histopathological examination remains the diagnostic gold standard.

In the present case report, the initial panoramic radiograph indicated expansile changes consistent with imaging features of ameloblastoma; however, subsequent tissue biopsy confirmed ACC. The lesion primarily involved the left maxillary alveolar process and body, forming a soft tissue mass measuring ~4.7x4.2 cm with expansile growth. This pattern suggests a tumor epicenter within the maxillary body. In contrast to these observations, tumors originating in the maxillary sinus with secondary maxillary invasion typically demonstrate eccentric

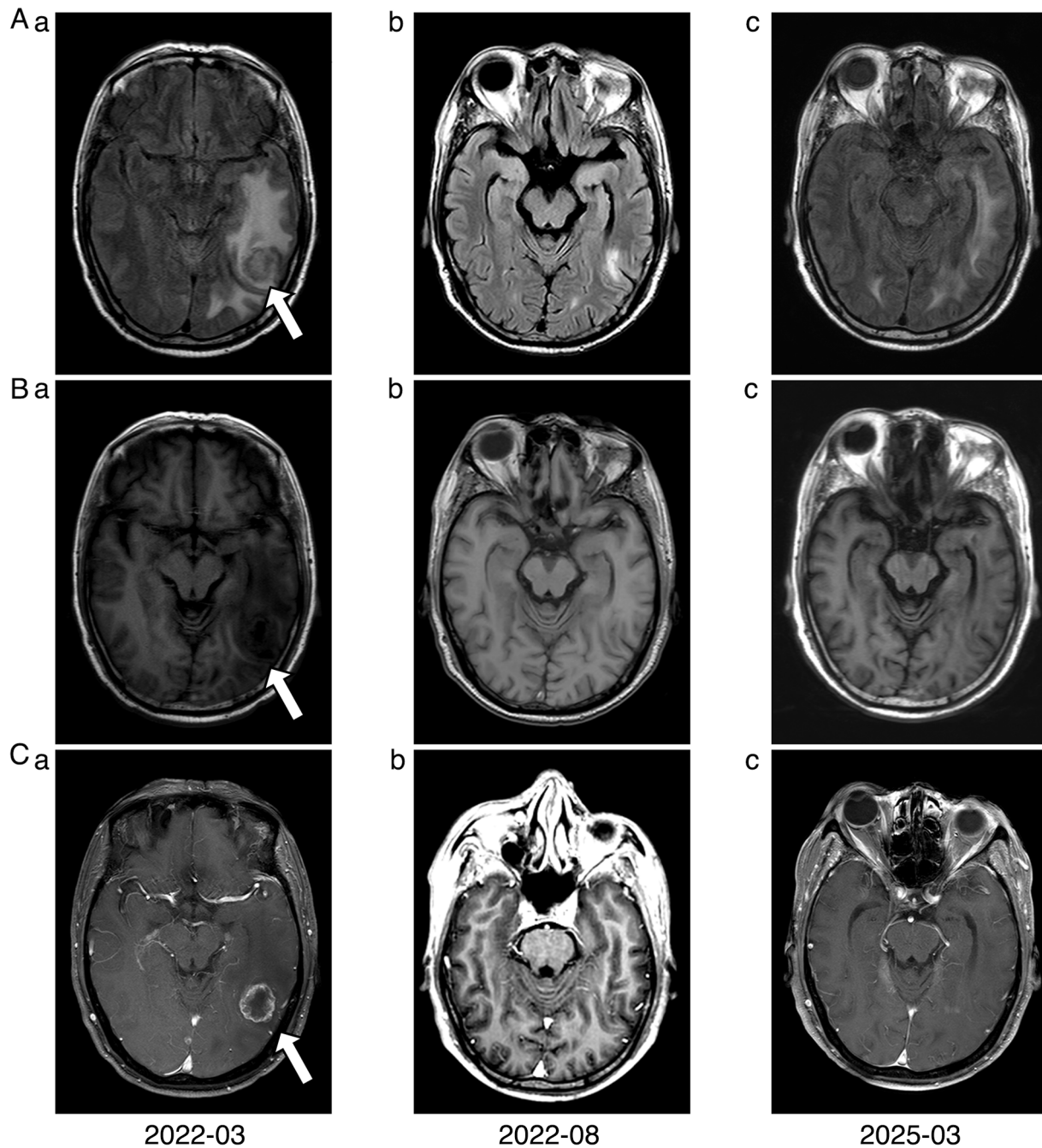


Figure 6. Follow-up brain MRI of the left occipital lobe metastasis. Initial MRI. (Aa) T2-FLAIR: 2.1x1.9 cm left occipital lobe mass with extensive perilesional edema (arrow). (Ba) T1WI: Mass isointense to slightly hypointense (arrow). (Ca) Contrast T1WI: Irregular rim enhancement (arrow). (Ab-Cb) August 2022: Lesion size reduced, enhancing rim thinned, and perilesional edema nearly resolved. (Ac-Cc) March 2025: Stable residual cavity, no significant change, no new nodules/edema. T1WI, T1-weighted imaging.

growth centered on the sinus cavity (16). Based on these findings, a diagnosis of primary ACC arising within the maxilla was proposed.

The standard treatment for ACC is radical surgery combined with postoperative radiotherapy, with a 5-year survival rate of ~75%. However, due to high recurrence and metastasis rates, 10 and 15-year survival rates drop to 50-60 and 30-35%, respectively (4). For recurrent or metastatic ACC, palliative chemotherapy is the mainstay, although outcomes are limited. This underscores the value of documenting novel strategies in challenging cases, as emphasized by Wáng (17) regarding the role of case reports in driving therapeutic innovation. The

present case, with its unprecedented metastatic pattern and sequential multimodal approach, contributes to this body of knowledge. The sequential strategy used for the present patient aligns with the framework by Zupancic *et al* (18). This approach extended overall survival to 68 months from initial diagnosis (January 2020) to last follow-up (September 2025).

Research on the cisplatin + nab-paclitaxel (TP) regimen in metastatic ACC is controversial (19), however, both drugs are recommended by the National Comprehensive Cancer Network guidelines (20). The majority of the studies support combination therapy over monotherapy, although data on maintenance chemotherapy are limited and inconclusive (5). The patient in

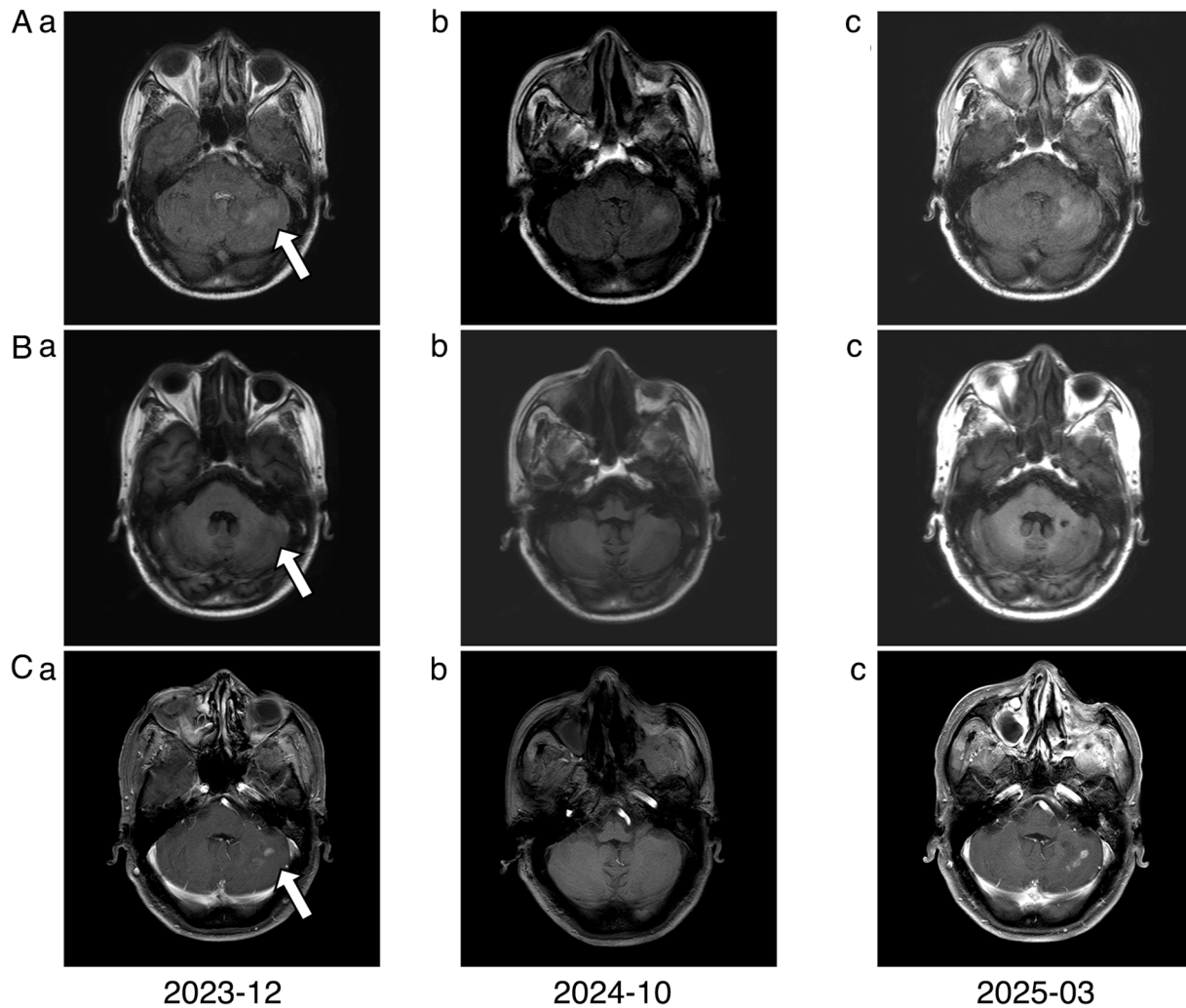


Figure 7. Follow-up brain MRI of the cerebellar metastasis. (Aa) T2WI/FLAIR: Patchy left cerebellar hemisphere hyperintensity (edema) with indistinct margins and mass effect (arrow). (Ba) T1WI: Slightly hypointense lesion (arrow). (Ca) Contrast T1WI: Irregular patchy lesion with peripheral-predominant (rim) heterogeneous enhancement (arrow). (Ab-Cb) October 2024: Marked lesion regression, decreased enhancement, near-complete edema resolution. (Ac-Cc) March 2025: Punctate/small patchy residual enhancement only, no new nodular enhancement or edema. T1WI, T1-weighted imaging.

the present study achieved sustained stabilization of pleural metastases following six cycles of TP chemotherapy supplemented with nab-paclitaxel maintenance therapy. This favorable outcome suggests that adding nab-paclitaxel maintenance following six cycles of TP chemotherapy may represent an effective treatment modality for pleural metastasis.

The patient developed brain metastasis despite stable extracranial disease, likely due to cisplatin and paclitaxel poorly penetrating the blood-brain barrier, allowing tumor cells to spread intracranially via perineural invasion. Notably, the patient presented with pleural metastasis first, which may have a higher risk for brain involvement, an area which requires further research.

Due to the propensity of ACC for perineural invasion and the ability of temozolomide to cross the blood-brain barrier, broad-spectrum antitumor activity demonstrates significant efficacy against the majority of intracranial malignancies (for example, gliomas) (21). With the consent of the patient, temozolomide (75 mg/m² orally, once daily) was administered orally daily. This regimen achieved favorable therapeutic outcomes without severe adverse reactions.

Lenvatinib, a multitargeted tyrosine kinase inhibitor, is recommended for the treatment of advanced ACC by multiple guidelines (20,22). Furthermore, studies have demonstrated the efficacy of tegafur in ACC treatment (23,24). Due to the current patient's intracranial disease progression, preserved performance status, high tumor burden and the anticipated limited efficacy of monotherapy, combined therapy with tegafur and lenvatinib was initiated under close monitoring for adverse events. This regimen resulted in stabilization of both intracranial and extracranial metastases for >1 year. These findings suggest that tegafur plus lenvatinib represents a viable treatment option for patients with multi-metastatic ACC experiencing localized progression following previous treatment with chemotherapy.

Overall, to the best of our knowledge, the present study reports the first documented case of maxillary ACC with sequential metastases to the pleura and intracranial sites. Following six cycles of TP regimen chemotherapy, maintenance therapy with nab-paclitaxel resulted in sustained significant control of pleural metastases. In addition, to the best of our knowledge, the present study represents the first application of temozolomide combined with nab-paclitaxel following brain metastasis radiotherapy,

yielding a favorable intracranial response. Furthermore, the tegafur-levatinib combination proved to be a viable regimen following second-line chemotherapy resistance in this patient. Whether this multimodal approach can be extended to other metastatic maxillary ACC cases warrants further investigation.

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Availability of data and materials

The data generated in the present study may be requested from the corresponding author.

Authors' contributions

BH and JS were responsible for clinical management of the patient, analyzed and interpreted clinical data, constructed figures, and drafting the manuscript. QYY was responsible for diagnostic analysis, performed imaging, advising on treatment, and manuscript revision. LZ was responsible for conceptualization, supervision, critical revision and final approval. BH and JS confirm the authenticity of all the raw data. All authors have read and approved the final manuscript.

Ethics approval and consent to participate

Written informed consent was obtained from the patient for treatment.

Patient consent for publication

The patient provided written informed consent for publication of de-identified clinical details and images.

Competing interests

The authors declare that they have no competing interests.

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