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Case Report

Very late intracranial extraparenchymal solitary metastasis of adenoid cystic carcinoma of the parotid gland: A case report and literature review

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ABSTRACT

Background: Adenoid cystic carcinoma (ACC) of the parotid gland often manifests as distant metastasis long after the initial surgery, resulting in a poor long-term prognosis. The most common sites of metastasis are the lungs, liver, and bones. Single intracranial metastasis is very rare.

Case Description: A 43-year-old woman with a history of surgical removal of ACC 24 years prior presented with Gerstmann syndrome and right hemianopsia. Head magnetic resonance imaging revealed a solitary extra-axial tumor in the left occipital region. The tumor was surgically removed and pathologically diagnosed as a metastasis of cribriform-type ACC. Since she had no other systemic metastasis, she did not receive adjuvant treatment and has remained recurrence-free for 35 months postoperatively.

Conclusion: Very late intracranial solitary metastasis of ACC is extremely rare. Due to the risk of delayed recurrence in pathologically confirmed cribriform type ACC, long-term follow-up is recommended.

Keywords: Adenoid cystic carcinoma, Cribriform, Metastasis, Surgery, c-KIT, CD56

INTRODUCTION

Adenoid cystic carcinoma (ACC) has an annual incidence of 3–4.5 cases/million people, accounting for 1–2% of all head and neck tumors and 10% of all parotid gland tumors.[3,9,13] While the short-term prognosis appears favorable, the 5- and 10-year survival rates for the primary tumor are approximately 60% and 50%, respectively. ACC proliferates slowly, leading to a high rate of late metastasis (33–50%) and a poor long-term prognosis, with a 20-year survival rate of around 20%.[3,4,12,14,15]

The most common sites of metastasis are the lungs, liver, and bones, with many cases of skull base metastasis reported due to direct or nerve invasion. Isolated intracranial metastasis occurring 15 or more years after the initial surgery is exceptionally rare.[2,6,10]

We herein report a case where a single intracranial extra-axial metastasis was detected 24 years after the surgical removal of ACC from the parotid gland. Few cases of late recurrence manifesting as isolated intracranial metastasis after ACC surgery have been reported.

CASE DESCRIPTION

The patient was a 43-year-old woman who had previously undergone surgery at 19 years of age to remove a tumor from her right parotid gland. Based on pathological findings, the patient was diagnosed with cribriform ACC and received postoperative radiation therapy. The patient experienced no recurrence for 10 years after the surgery and completed her outpatient follow-up. Twenty-four years post-surgery, the patient complained of headaches. Head magnetic resonance imaging (MRI) revealed an extra-axial tumor in the left occipital region, for which the patient was referred to our hospital. On the first visit, the patient was alert and displayed alexia, agraphia, and right homonymous hemianopia. Head MRI showed a low signal on T1-weighted imaging (T1WI), a high signal on T2-weighted imaging, and a low signal on diffusion-weighted imaging. Gadolinium-enhanced T1WI showed a lesion approximately 6 cm in diameter with rounded edges and uneven internal contrast. Head computed tomography (CT) revealed osteolytic changes in the skull and no intra-tumoral calcification [Figure 1].

The patient underwent surgical tumor resection under general anesthesia. In the prone position, a hockey stick skin incision was made. Skin flap eversion revealed osteolytic changes in the skull and a tumor extending into the periosteum. The tumor was extended beneath the dura mater, and the dura mater was partially defected. The tumor was white and soft, with pial invasion, and was completely removed macroscopically. The dura mater was partially defected and reconstructed using GORE-TEX (W.L. Gore and Associates, Newark, DE, USA). The bone was removed to create a sufficient margin around the infiltrated area, and a titanium mesh plate was used to fill the resulting bone defect. The patient recovered from general anesthesia without neurological deterioration. Pathological diagnosis confirmed ACC featuring a predominantly cribriform pattern and minor tubular components. Immunostaining indicated marginal positivity for Ki-67, c-KIT, and CD56, with a notable absence of CD56-positive cells in the initial surgery [Figure 2].

Postoperative course

The patient recovered well after the surgery without complications. Postoperatively, the hemianopsia and Gerstmann syndrome improved. Head MRI with gadolinium enhancement showed no residual tumor [Figure 3]. 18F-fluorodeoxyglucose positron emission tomography-

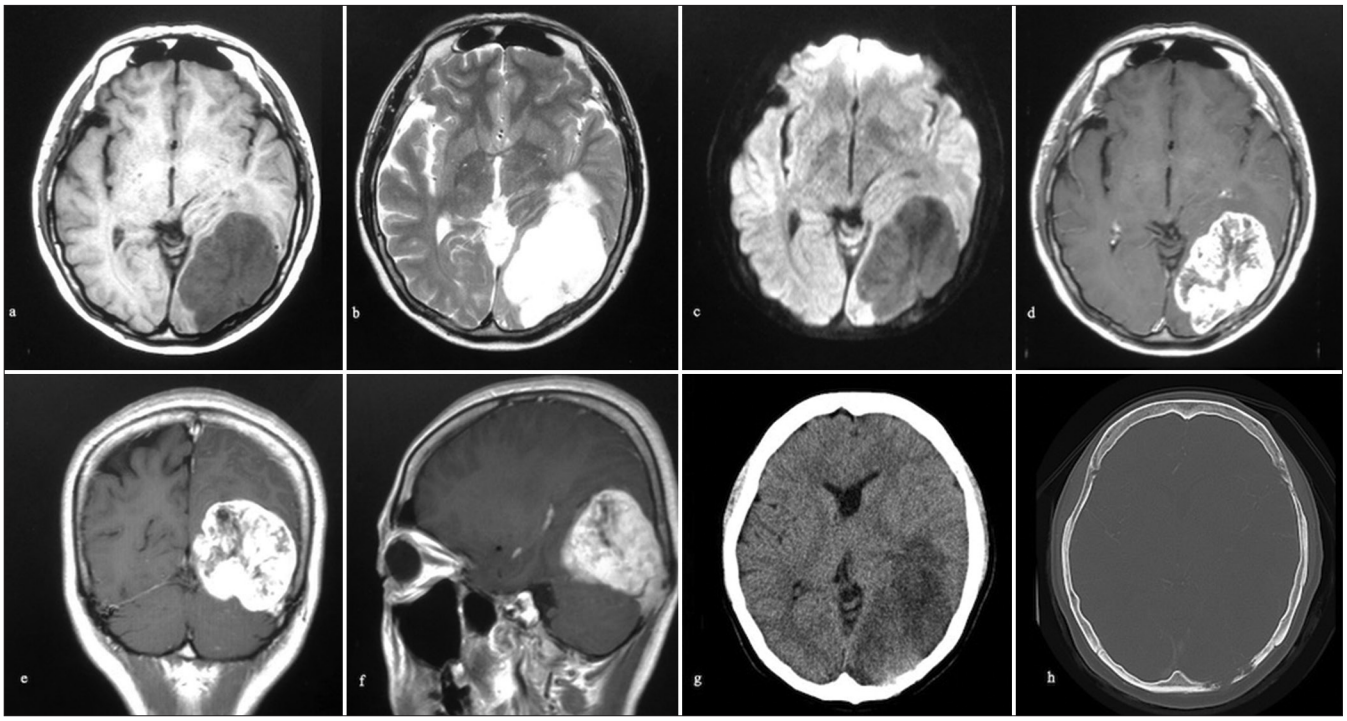

Figure 1: (a-c) Preoperative magnetic resonance imaging (MRI) T1-weighted imaging, T2-weighted imaging, and diffusion-weighted images showed a well-defined round tumor in the left occipital lesion. (d-f) Gadolinium-weighted images of the head MRI showed an irregularly enhancing tumor. (g and h) Preoperative computed tomography of the head showed osteolytic changes in the cranium and no intratumoral calcification.

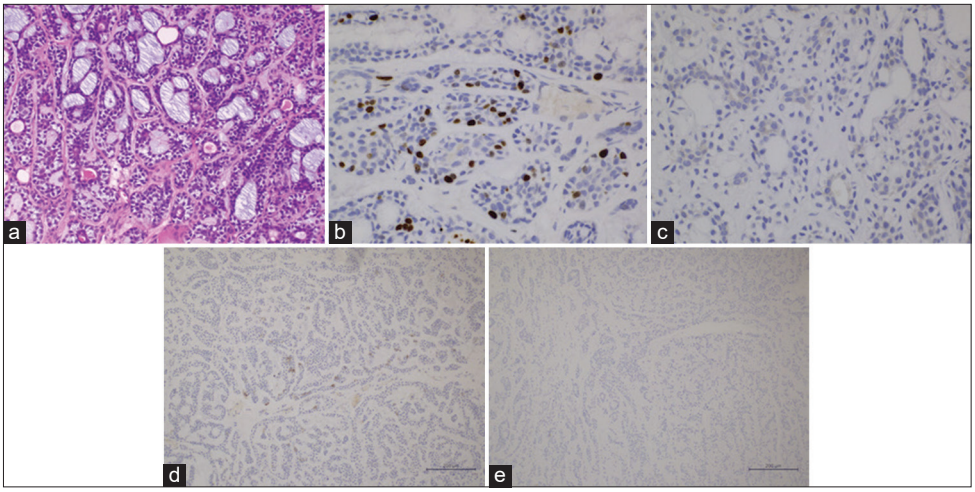

Figure 2: (a) The pathological diagnosis of the tumor with hematoxylin and eosin staining was adenoid cystic carcinoma, cribriform type. (b) Immunostaining for Ki67 was slightly positive. (c) Immunostaining for the c-kit was partially positive. (d) The immunostaining for CD56 in the brain metastasis was partially positive compared to the initial surgery. (e) The immunostaining for CD56 of the parotid gland in the initial surgery was negative.

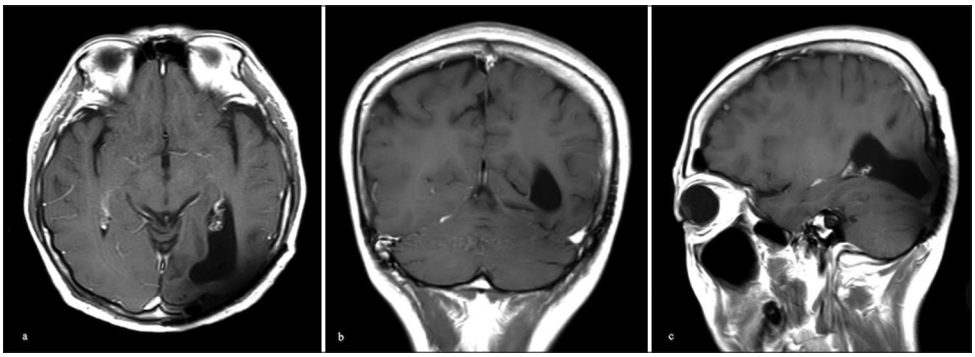

Figure 3: (a-c) Postoperative gadolinium-enhanced head magnetic resonance imaging showed no residual tumor.

CT showed no recurrence of the primary tumor or other metastatic lesions. The patient has remained recurrence-free for 35 months postoperatively.

DISCUSSION

This case illustrates the characteristics of ACC, particularly its capacity for significant delays in metastasis, challenging conventional management paradigms. Although ACC grows slowly, distant metastases may occur even with good primary site control. The reported distant metastasis rate is 8–60%,[3,11,15] and approximately 30% of patients with distant metastases have no recurrence of the primary tumor. One metastatic case reportedly occurred > 20 years after the first surgery.[10] Common metastases occur in the lungs, liver, and bones, yet the rarity of isolated brain metastasis calls for heightened awareness during differential diagnosis when confronted with isolated intracranial tumors. While the optimal follow-up period is difficult to determine, long-term follow-up is preferable.

Only seven cases of isolated intracranial metastases have been reported, including the present case.[1,2,5,7-9] Among these, two patients had intra-axial metastases, one had pituitary metastases, and four had intracranial extra-axial metastases. In the preoperative diagnosis of intracranial extra-axial brain tumors, the differential diagnoses included primary brain tumors like meningiomas and metastatic tumors. Some metastatic tumors may exhibit a dural-tail sign resembling meningiomas. However, the preoperative diagnosis is complex, and with a history of ACC, metastasis should be considered in the differential diagnosis.

The pathological diagnoses of these seven reported cases included five cases of the cribriform type and one each of the tubular and solid types. Three cases of isolated intracranial recurrence occurring > 15 years after the initial surgery

have been reported, including the present case. All three cases were of the cribriform ACC type.[2,10] Typically, the solid form is more malignant, leading to early and frequent metastases and poorer outcomes. Although the cribriform type is less aggressive than the solid type, it can still metastasize after a long period. While there is no consensus on the significance of immunostaining, c-KIT (CD117) and neural cell adhesion molecule (N-CAM, CD56) are generally considered growth factors. In our case, immunostaining was mildly positive for Ki-67, negative for c-KIT, and mildly positive for N-CAM (CD-56), although pathological examination of N-CAM in the initial surgery was negative. While the roles of these markers are debated, CD56's presence is notable as recent studies link it with metastatic potential and recurrence prognostic value in multiple cancers, including ACC.

At present, there is no standard treatment protocol for patients with metastatic ACC, with treatments tailored to individual patient profiles. Surgical resection and radiation therapy are typically preferred for solitary, operable tumors due to their effectiveness in suppressing tumor regrowth.[2] Recent advancements have introduced the possibility of molecular targeted therapy and immune checkpoint inhibitors in addition to classical chemotherapy.[12,10] Furthermore, the potential for tailored gene therapy offers promise for better recurrence control. This discussion reaffirms the complexity of ACC's metastatic behaviors and the need for long-term surveillance. It highlights the necessity for flexible treatment approaches, emphasizing personalized medicine as the optimal path forward in addressing the challenges presented by this enigmatic carcinoma.

CONCLUSION

We presented a case of solitary distant intracranial metastasis of ACC of the parotid gland 24 years after the initial surgery. ACC may metastasize solely within the cranium after a long period without local postoperative recurrence. The prognosis for cases like these can be favorable following complete surgical resection. While the effectiveness of adjunctive treatments such as radiation or chemotherapy remains uncertain, this case highlights the necessity for ongoing research into new therapies, including molecular target therapy.

Ethical approval

The Institutional Review Board approval is not required.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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