

Metastatic versus metachronous adenoid cystic carcinoma in the lacrimal gland fossa: a case report

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Abstract

A 53-year-old woman presented with a left painless enlarging lacrimal fossa lesion with hypoglobus and choroidal folds. 18 months earlier, she had undergone surgery and chemoradiotherapy for right maxillary sinus adenoid cystic carcinoma. After initial 1.5T magnetic resonance imaging, a double primary of left lacrimal gland adenoid cystic carcinoma with tricompartimental (frontal bone, lacrimal and temporalis fossa) involvement was suspected. However, subsequent high-field multiparametric 3T magnetic resonance imaging suggested a solitary metastasis with a subperiosteal location and an uninvolved lacrimal gland. The solitary metastasis was confirmed intraoperatively and pathologically.

Key words: Carcinoma, adenoid cystic; Lacrimal apparatus; Multiparametric magnetic resonance imaging

Introduction

Adenoid cystic carcinoma (ACC) is the most common lacrimal gland epithelial cancer, accounting for up to 60% of all cases in one series.¹ Primary tumor excision, exenteration and/or craniofacial resection together with intra-arterial chemotherapy and external beam irradiation are treatment options depending on tumor staging and clinician experience.^{2,3} We herein describe a patient who presented

with an expanding solitary subperiosteal metastasis of maxillary sinus ACC mimicking a double primary of contralateral lacrimal gland ACC.

Case presentation

In December 2019, a 53-year-old woman presented to our hospital with worsening of the left lower visual field loss. In January 2018, she had undergone subtotal maxillectomy, free fibular flap reconstruction, and adjuvant chemoradiotherapy elsewhere for a pT3N0 ACC of the right maxilla. On examination, her left eye had 7-mm proptosis, 3-mm hypoglobus, limited elevation and superotemporal choroidal folds, and a vaguely palpable non-tender swelling over the lacrimal gland fossa. 1.5T magnetic resonance imaging (MRI) of the orbit showed a well-circumscribed, homogeneously contrast-enhancing, T1-hyperintense lesion measuring 2.0×1.7×0.8 cm over the left lacrimal gland fossa, with frontal bone involvement and suspicious contiguous lymphadenopathy (**Figure**).

In March 2020, 3T MRI showed a 2.4×2.4×1.7 cm subperiosteally located lesion in the lacrimal gland fossa indenting onto the left globe with interval increase in size. It was slightly T1-hyperintense to the extraocular muscle and lacrimal gland, heterogeneous T2-hyperintense with small cystic component and a higher degree of contrast enhancement. A normal looking lacrimal gland was inferiorly displaced. Bony dehiscence and extension of the lesion into the temporalis fossa was demonstrated (**Figure**). Positron emission tomography showed additional multiple nodules in bilateral lungs.

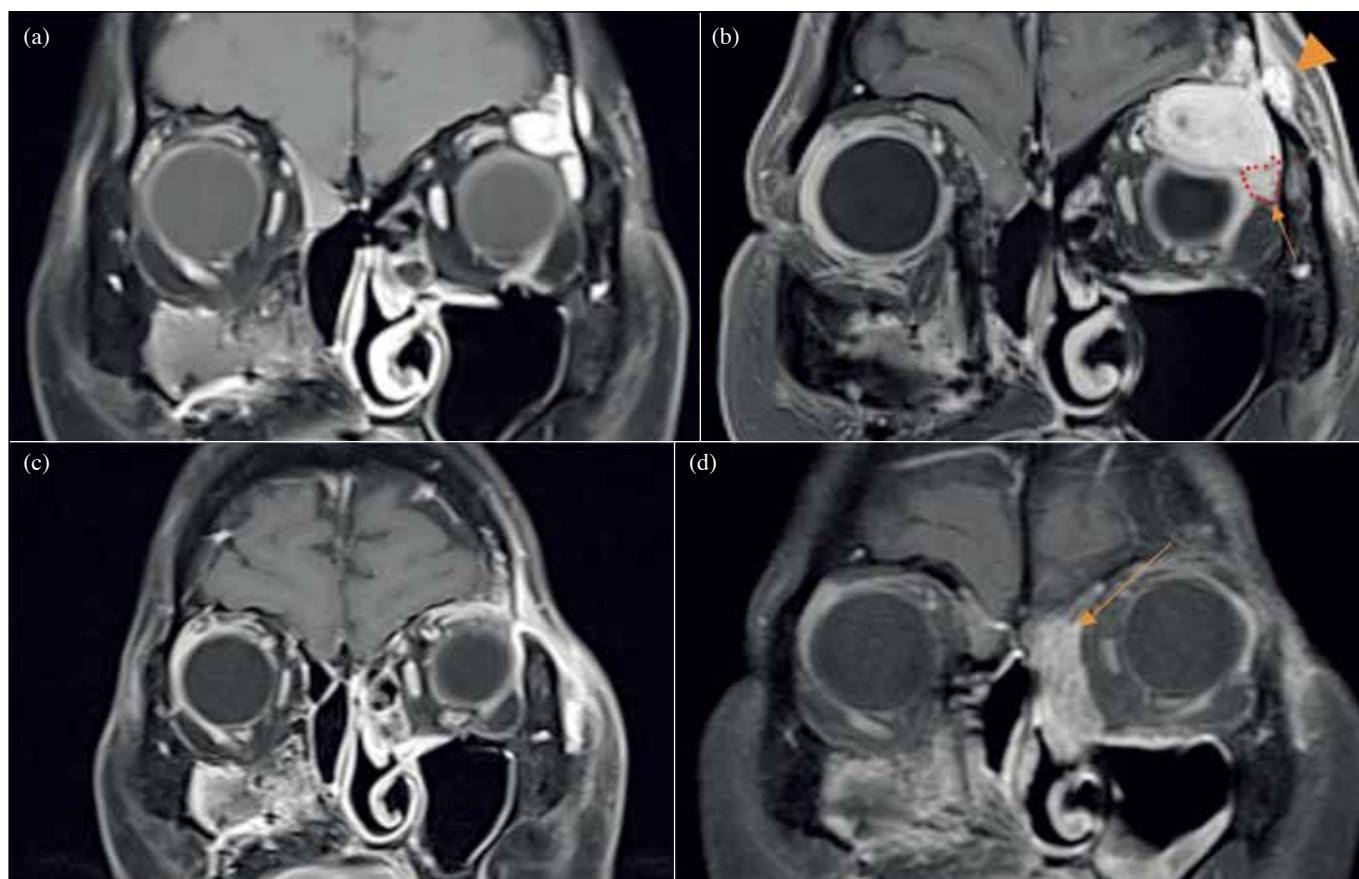


Figure. Post-contrast T1-weighted fat-suppressed coronal magnetic resonance imaging (MRI) of the orbit: (a) In December 2019, 1.5T MRI showing a homogeneously contrast-enhancing lesion suspicious of lacrimal fossa tumor over the left lacrimal gland fossa with frontal bone involvement. (b) In March 2020, 3T MRI showing the enlarging lesion and ultra-fine anatomical details, including its subperiosteal location, a small cystic (necrotic) component, normal-looking lacrimal gland displaced inferiorly (arrow), and bony invasion at the frontozygomatic suture extending into the temporalis fossa (arrowhead). (c) In July 2020, 1.5T MRI showing the reduced subperiosteal lesion at 3 months after surgical excision. (d) In December 2020, 1.5T MRI showing no recurrence over the lacrimal fossa at postoperative 7 months, but new enhancing soft tissue (arrow) over the left medial orbit suspicious of metastatic deposit.

In March 2020, the patient underwent left subbrow orbitotomy and excisional biopsy. The subperiosteal location of the tumor, the frontozygomatic bone defect, and normal lacrimal gland were confirmed intraoperatively. She received postoperative adjuvant radiotherapy. In July 2020 and November 2020, postoperative MRI showed no recurrent metastatic disease but suspicious growth over the medial orbit or lacrimal sac region (**Figure**).

Discussion

This case illustrates the importance of superior anatomical delineation of complex orbital lesions using high-field 3T MRI, which provides valuable roadmap for pre-operative planning. Multiparametric MRI involves diffusion and perfusion-weighted sequences and provides valuable functional information in addition to anatomical details.⁴ The additional use of diffusion-weighted imaging significantly improves the differentiation of benign from malignant orbital masses.⁵ Substantial enlargement of a noninflammatory

lacrimal fossa lesion within a short period together with invasion to the surrounding frontal bone and temporalis fossa was highly suggestive of malignancy. However, on high-field 3T MRI, the subperiosteal location and the normal lacrimal gland concluded that the diagnosis of an advanced primary lacrimal gland ACC was unlikely.

Only one case of metachronous primary ACC has been reported; the patient had a history of an index maxillary sinus ACC with esophageal carcinoma 14 years later.⁶ Whereas there have been 12 cases of lacrimal gland or maxillary sinus ACC with metastasis to other organs including liver, kidneys, and lungs.⁷⁻¹⁶ Therefore, primary versus metachronous ACC are differential diagnoses. High-field multiparametric MRI is an important tool to assess orbital masses.

Author contributions:

Concept or design: KKLC

Acquisition of data: KKLC, WCWC

Analysis or interpretation of data: PPYW, KKLC
 Drafting of the article: PPYW
 Critical revision for important intellectual content: KKWC & KKLC, WCWC
 All authors had full access to the data, contributed to the study, approved the final version for publication, and take responsibility for its accuracy and integrity.

Conflicts of interest

The authors have no conflicts of interest to disclose.

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Patient consent

The patient was treated in accordance with the Declaration of Helsinki. The patient provided informed consent for the treatment/procedures. The patient provided consent for publication.

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