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Case Report: Unexpected Dural Metastasis in a Patient with Adenoid Cystic Carcinoma of the Lacrimal Gland Four Years After Surgery

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ABSTRACT

Adenoid cystic carcinoma (ACC) of the lacrimal gland is a rare, slow-growing malignancy with a tendency for late recurrence and distant metastasis. Despite successful initial treatment, ACC can recur years later, often in a clinically silent manner. This case report describes a 45-year-old patient with a history of ACC of the lacrimal gland, who underwent left eye exenteration followed by radiotherapy. Four years after the initial treatment, a routine MRI revealed an unexpected dural metastasis in the left frontotemporal region. The mass demonstrated pachymeningeal thickening and enhancement, indicative of malignant spread. Remarkably, the patient remained asymptomatic, underscoring the importance of long-term follow-up and imaging surveillance in detecting late recurrences and metastases. This case highlights the unpredictable nature of ACC, emphasizing the need for vigilant monitoring even years after successful treatment.

KEYWORDS :

Adenoid cystic carcinoma, lacrimal gland, metastasis

MAIN ARTICLE

INTRODUCTION

Adenoid cystic carcinoma (ACC) is a rare and aggressive malignancy that most commonly affects the salivary glands but can also arise in other structures, including the lacrimal gland. It is characterized by a slow yet relentless growth pattern, often presenting with late-stage recurrence or metastasis. Despite its indolent behavior, ACC has a propensity for local invasion and distant spread, which makes early detection crucial for successful management. The lacrimal gland, though infrequently affected, is a site where ACC can cause significant morbidity due to its proximity to vital ocular and neural structures.

The typical treatment for lacrimal gland ACC involves surgical resection, which may include exenteration of the orbit, followed by adjuvant radiotherapy to reduce the risk of recurrence. However, recurrence can occur years after initial treatment, often in asymptomatic patients. Metastasis, including dural involvement, remains an uncommon but severe complication. This case report presents an unexpected dural metastasis from ACC of the lacrimal gland in a patient who had been free of disease for four years, underscoring the need for continued surveillance and the potential for late metastatic spread, even in the absence of clinical symptoms.

CASE REPORT :

The patient, a 45-year-old with no significant medical or surgical history, was initially diagnosed with adenoid cystic carcinoma of the right lacrimal gland, for which he underwent left eye exenteration followed by radiotherapy at the National Oncology Institute. Post-surgery, he had regular follow-up MRIs, with the most recent one, conducted about one to two years after treatment, showing no signs of tumor recurrence and a clear left orbital cavity. However, during a subsequent MRI, a new polylobulated extra-axial mass was discovered in the left frontal region, which exhibited an intermediate signal on T2-weighted imaging and intense, homogeneous enhancement after contrast administration **(Figure 2),** suggesting a malignant process. Further evaluation revealed bony infiltration in the frontal area, as well as pachymeningeal thickening and enhancement in the frontotemporal region **(Figure 3),** indicating possible dural involvement and a recurrence with local spread. Despite these concerning radiological findings, the patient remained asymptomatic, without any clinical signs such as pain, neurological deficits, or other functional symptoms, underscoring the importance of long-term imaging surveillance in detecting late recurrences even when the patient is clinically stable.

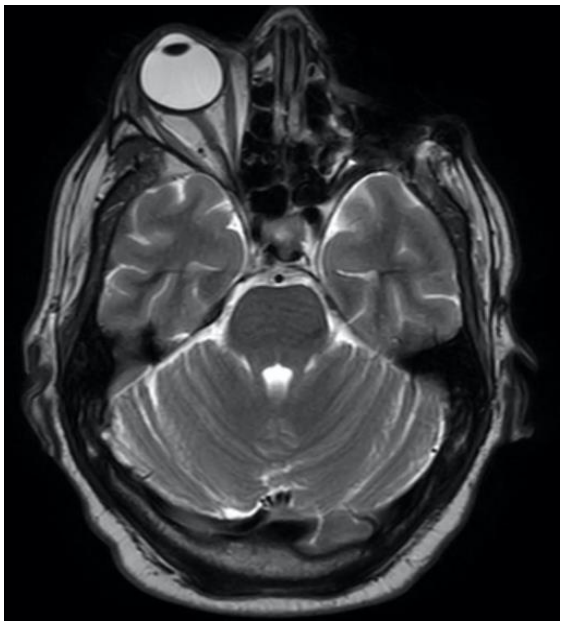

Figure 1 : *Axial section of a brain MRI in T2 sequence showing an empty left orbital cavity.*

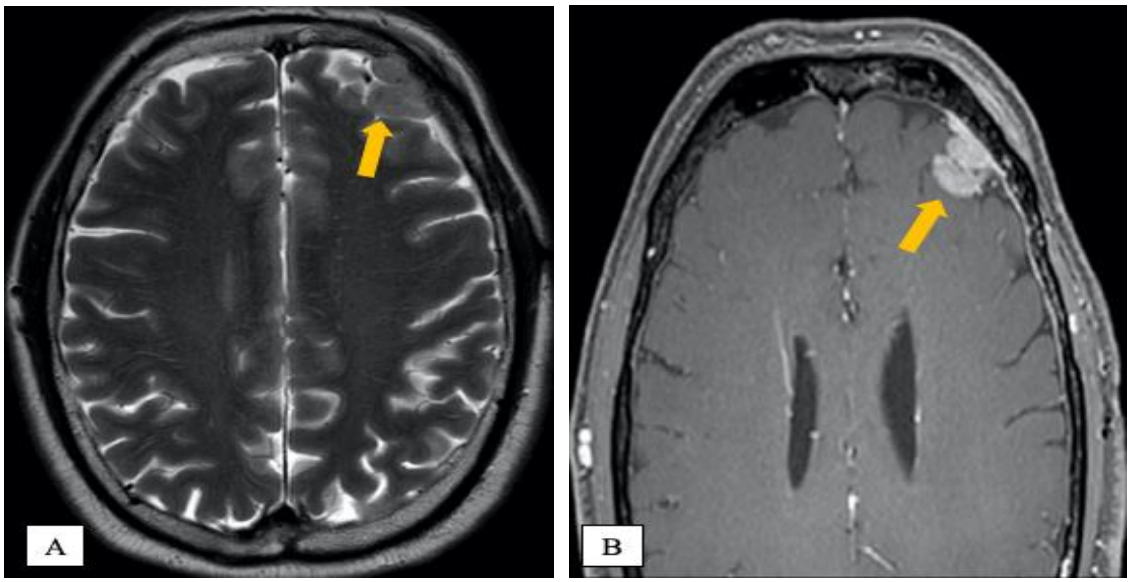

Figure 2 : *Brain MRI in axial sections: showing a polylobulated extra-axial mass in the left frontal region (ORANGE ARROW), which exhibited an intermediate signal on T2-weighted imaging (A) and intense, homogeneous enhancement after contrast administration (B).*

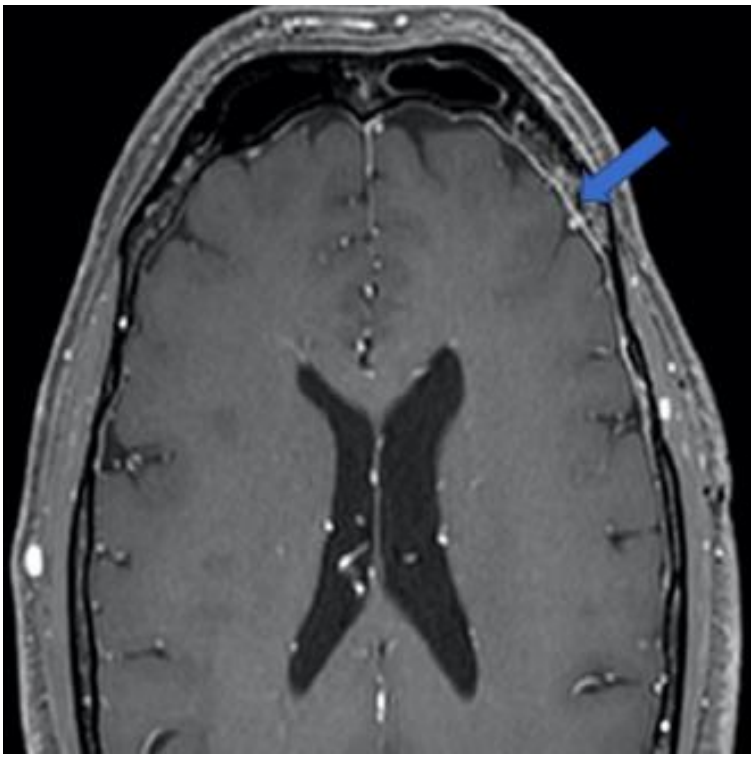

Figure 3: Axial brain MRI showing pachymeningeal thickening and enhancement in the left frontotemporal region (Blue arrow), indicating malignant involvement.

DISCUSSION :

Adenoid cystic carcinoma (ACC) is a rare malignant tumour that most commonly arises in the salivary glands. It is typically slow-growing but exhibits aggressive behaviour, characterised by a high propensity for late recurrences and distant metastases, which may occur several years after the initial diagnosis. ACC may also develop in other secretory glands[1][2].

It represents the most common malignant epithelial tumour of the lacrimal gland, although it remains rare overall. Despite its indolent growth, it is highly aggressive, with frequent local recurrence, bone and perineural invasion, distant metastases, and a high mortality rate despite treatment[3].

ACC accounts for approximately 1.6% of orbital tumours and constitutes the majority of malignant epithelial tumours of the lacrimal gland[4], It predominantly affects young to

middle-aged adults, with a peak incidence around 40 years of age, although cases in children have also been reported [5].

Clinically, patients typically present with exophthalmos, which is often non-axial, associated with pain (a feature suggestive of malignancy), ptosis, diplopia, or decreased visual acuity due to optic nerve compression[6].

Imaging—primarily based on magnetic resonance imaging (MRI)—typically demonstrates a solid mass in the superotemporal orbit, often associated with bone erosion or destruction and occasionally calcifications. Notably, the lesion may appear deceptively well circumscribed[1].

The mainstay of treatment is surgical, consisting of wide excision such as orbitectomy or orbital exenteration in cases of globe involvement, followed by adjuvant therapy based on histopathological findings[7]. More recently, neoadjuvant intra-arterial chemotherapy combined with surgery has shown improved survival outcomes compared with conventional approaches in selected studies [8].

Despite appropriate management, both local and distant recurrences remain frequent[9], Intracranial extension or metastasis may occur through direct invasion, perineural spread, or haematogenous dissemination, although such occurrences are relatively uncommon. Local extension and perineural spread commonly involve the skull base, anterior and middle cranial fossae, cavernous sinus, dura mater, and occasionally the brain parenchyma. Tumour spread may also follow cranial nerves toward the cavernous sinus, temporal bone, cerebellopontine angle, convexity dura, and tentorium cerebelli [10].

Brain involvement may also occur via haematogenous spread, resulting in multiple meningeal metastases. In a series of 39 patients, brain involvement was observed in 61.5% of those with distant metastases, with a frequency comparable to pulmonary metastases[11]. In general, dural metastases are thought to arise via either haematogenous or perineural routes.

Management of brain metastases is primarily surgical in cases of solitary lesions, often combined with radiotherapy or stereotactic radiosurgery for meningeal involvement or multiple small lesions. In one reported case with a two-year follow-up, this approach resulted in complete local control and preservation of quality of life[12].

CONCLUSION :

ACC of the lacrimal gland can lead to brain metastases or intracranial spread, often via perineural and bone invasion of the skull base, but also via the haematogenous route. The brain and dura mater are common sites among documented distant metastases, and focal techniques such as radiosurgery can control certain selected lesions.

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The authors declare that they have no conflicts of interest.

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