Case Report

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Epithelial-Myoepithelial Carcinoma of the Lacrymal Gland

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Abstract

Epithelial-myoepithelial carcinoma is a very uncommon tumor of the orbit, apparently behaves like a low-grade malignancy and is associated with good survival rates. However, a good prognosis for survival is tentative at best, in part because of the rarity of the lesion. Herein, we report an EMC of the lacrimal gland in a 95-years-old woman and review the available literature on this very rare tumor.

Keywords: Epithelial-myoepithelial; Carcinoma; Lacrymal gland; EMC

Introduction

In all intrinsic lacrimal gland masses, 28% are epithelial neoplasms. Epithelial-Myoepithelial Carcinoma (EMC) and Myoepithelial Carcinoma (MC) are uncommon epithelial malignancies accounting for 1% of all salivary glands that have been rarely reported in the lacrimal gland [1]. Epithelial-Myoepithelial Carcinoma (EMC) is a malignant tumor characterized by biphasic morphology of duct-like structures surrounded by clear myoepithelial-like cells. While EMC in the salivary gland is known to behave as a low-grade malignancy associated with good survival rates of approximately 94% and recurrences are reported even decades after initial surgery, its behavior in the lacrimal gland remains to be clarified.

Case Study

Here in, we report an EMC of the lacrimal gland in a 95-years-old woman and review the available literature on this very rare tumor.

This 95-year-old woman presented with history of a right ocular pain of 1 year duration with rapid decreased visual acuity. Examination showed a blepharitis, chemosis and purulent secretions of the right eye [2-5]. On palpation, a firm, rubbery, nontender mass was palpable through the upper eyelid. No significant comorbidity was present. Computed tomography scan showed a homogenously enhancing lobulated mass of the inner part of the right eyeball. It is responsible for deformation and loss of sphericity of right eyeball with very likely damage to the right inner muscle, the upper eyelid lifter and the small right-sided muscle **(Figure 1)**.

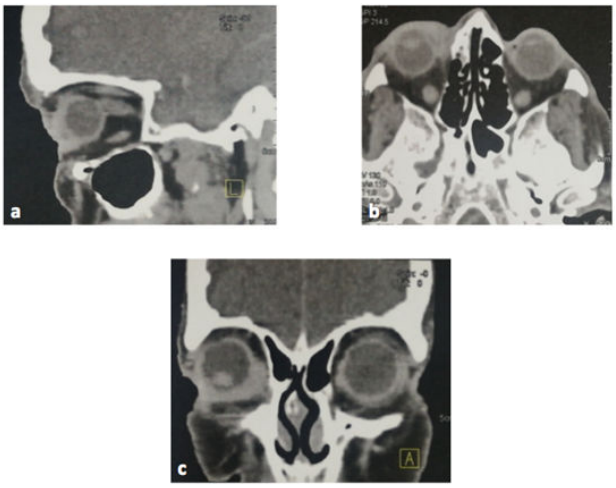

Figure 1: a) Coronal reconstruction; b) Axial plan; c) Sagittal reconstruction.

Based on clinical and radiologic findings, diagnosis of a lacrimal gland tumor was entertained and complete excision of the tumor was done through orbital exenteration.

Anatomopathological study showed a malignant tumor process with fusiform cells measuring (3.5 × 2.5 × 1.5) cm adhering to the globe ocular and optic nerve.

In immunohistochemical profile, the ductal epithelial cells were found to be strongly reactive for pan-cytokeratin AE1/AE3 for S100, AML and EMA.

A final diagnosis of epithelial-myoepithelial carcinoma was made. The patient had a local recurrence 3 months after surgery. Thoraco-abdomino-pelvic CT scan showed no evidence of metastasis [6]. The patient received palliative radiation therapy due to altered general condition, 13 Gy in 2 fractions of 6.5 Gy. She died the month after.

Epithelial-myoepithelial carcinoma is a rare neoplasm accounting for approximately 1% of all salivary gland neoplasms, with most cases arising in the parotid gland [7]. The similar morphology and clinical characteristics of salivary and lacrimal

gland tumors have led to the adoption of the same classification system for both neoplasms.

Therefore, the use of a classification system based on diverse histologically patterns as used for salivary gland tumors can also be successfully applied to the study of epithelial lacrimal gland tumors and has expanded the comprehension of the lacrimal gland tumor behavior.

Discussion

In the salivary glands, EMC is considered a low-grade malignancy, with a recurrence rate of 36.3% and a 5-year survival rate of 93.5%.

EMC can appear de novo or from malignant transformation of a benign tumor. Malignant tumor may mimic a benign lesion by deceptively appearing as a painless and slowly growing mass.

The first report was in 1994 of a 63-year-old man who presented with slowly progressive, painless proptosis for 8 years with sudden increase in size over 2 months, possibly heralding malignant change. Histopathology of the tumor showed features classical for EMC with areas of benign mixed tumor at the periphery [8]. The authors proposed that the malignant tumor had arisen from the preexisting pleomorphic adenoma.

Theoretically, computed tomographic scanning with contrast could help to discriminate epithelial from lymphoid or inflammatory lesions of the lacrimal gland. Radiologic features such as hyperostosis and tumor calcification, which were generally considered to be features of malignancy, might not be apparent in all cases. Theoretically, computed tomographic scanning with contrast could help to discriminate epithelial from lymphoid or inflammatory lesions of the lacrimal gland [9-13]. Pathological diagnosis by open biopsy is therefore the last resort in confirming underlying disease.

Recurrence and metastasis rates of epithelial-myoepithelial carcinoma from salivary gland have been reported to be from 35% to 50% and from 8.1% to 25%, respectively. The presence of tumor at the surgical margin suggests that radiotherapy or orbital exenteration may be necessary. In a report case of Wiwatwongwana, Surgical treatment was followed by adjuvant radiotherapy of 6000 cGy because of positive margins.

Conclusion

This rare tumor may behave as a low-grade malignant neoplasm, however, lifelong follow-up is required as recurrence or metastasis may occur years after surgery. Epithelial myoepithelial carcinoma of the salivary gland is considered to be a low grade malignancy, with a recurrence rate of 36.3% and a survival rate of 93.5% and 81.8% at 5 and 10 years. Factors significantly affecting disease free survival were positive margins, lymphovascular invasion, necrosis, and myoepithelial anaplasia.

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