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Sir,

Oncocytic adenocarcinoma of the lacrimal gland: an unusual presentation

We report a case of lacrimal gland oncocytic adenocarcinoma (OCA) whose initial presentation mimicked that of an isolated sixth nerve palsy rather than neoplastic tumour.

Case Report

A 76-year-old male was referred with diplopia and proptosis. He had been under review at his local hospital for an isolated sixth nerve palsy that had been well controlled with prismatic correction. His past medical history was unremarkable, with no history of diabetes, hypertension, or deranged thyroid function.

On right ocular examination, best-corrected visual acuity was 6/6 and optic nerve function was normal. A palpable mass was noted superotemporally, with a 2 mm proptosis. Slit lamp examination was unremarkable. MRI showed an enhancing extraconal mass involving the lacrimal gland and abutting the globe (Figure 1). There was no intracranial extension and systemic evaluation showed no metastasis.

An anterior orbitotomy was performed and the tumour was excised en-bloc. The tumour appeared as a solid, craggy lesion without attachments to lateral rectus muscle. Histolopathological examination demonstrated fibrous tissue and fat extensively infiltrated by carcinoma with multiple perineural, vascular, and perivascular invasion. The cells had oncocytic cytoplasm, enlarged nuclei, and numerous mitotic bodies. These features were consistent with OCA (Figure 2). Unfortunately,

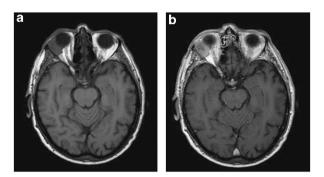


Figure 1 (a) T1 non-enhanced axial MR image showing an extraconal mass in the right orbit involving the lacrimal gland. There was no evidence of intracranial involvement. (b) T1 post-contrast axial MR image.

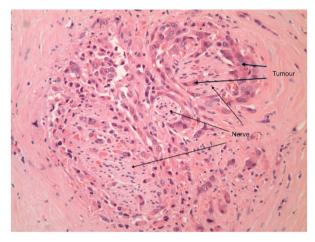


Figure 2 Histopathology slide showing an invasive OCA with perivascular and perineural invasion.

despite en-bloc removal of tumour, histological margins were not clear. Repeat CT scan showed residual tumour. The case was considered at a multidisciplinary meeting with exenteration being recommended.

Comment

OCA is a malignant epithelial tumour arising in the ductal cell lining of apocrine glandular structures.¹ Prognosis is poor as it is a high-grade neoplasm with infiltrative growth pattern and tendency to recur and metastasize. OCA may involve the caruncle, the conjunctiva, the lacrimal sac, and more rarely, the lacrimal gland. To date, only four cases of lacrimal gland OCA have been reported.^{2–5}

One possible explanation as to why the presumed diagnosis of sixth nerve palsy was made initially instead of mechanical limitation, and therefore not prompting any further investigations, might be early direct neural invasion with no evident mass effect. Our patient has remained disease-free for 24 months, but careful followup is recommended as metastasis appears to be the most important prognostic factor. OCA should be considered in the differential diagnosis of lacrimal gland lesions and exenteration is the treatment of choice.

Conflict of interest

The authors declare no conflict of interest.

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Sir,

Rapid corneal adrenochrome deposition from topical ibopamine in the setting of infectious keratitis

Owing to its ability to act as both a direct and indirect sympathomimetic agonist, topical ibopamine has been used to treat chronic refractory hyptotony.¹ Ibopamine is rapidly hydrolyzed by corneal esterases to epinine, which then undergoes the same *in vivo* oxidation process as epinephrine to form adrenochromes. In acidic mediums, ischemic environments, and in the presence of reactive oxygen species adrenochrome becomes unstable and can undergo autooxidation to its melanin degradation products.^{2,3} Although corneal pigmentation from topical epinephrine has been observed,⁴ and such depositions have been reported on a Boston keratoprosthesis with the use of ibopamine,⁵ such deposition from ibopamine has not, to our knowledge, been reported in the cornea itself.

Case report

A 50-year-old HIV-positive man underwent a penetrating keratoplasty in his left eye for a failed endothelial graft in the setting of significant anterior stromal scarring. He had a complex ocular history including cytomegalovirus retinitis status post ganciclovir intravitreal implants bilaterally and immune reconstitution uveitis status post flucinolone intravitreal implants bilaterally. He also had chronic hypotony in both eyes for which he was started on ibopamine 2% eye drops three times daily (compounded at Leiter's Pharmacy, San Jose, CA, USA) 3 months before the penetrating keratoplasty. Pre-operatively, the ibopamine use resulted in an increase in intraocular pressure from 5 to 7 mm Hg. After surgery, no epithelial defect was seen, and in addition to the ibopamine eye drops, routine topical antibiotic and corticosteroid coverage with polymixin B-trimethoprim and prednisolone acetate 1%, respectively, each four times daily was given. A peripheral corneal epithelial defect was seen 1 week post-operatively, and the defect persisted despite aggressive lubrication and tapering of his post-operative corticosteroid eve drops. Cultures were not taken at this point, as there was no clinically obvious infiltrate present.

Five weeks post-operatively, a brown-pigmented deposition appeared within the margins of the epithelial defect in the nasal peripheral corneal graft (Figure 1). The pigmentation was concentrated as a plaque overlying the de-epithelialized cornea and extended diffusely into the anterior one-third of the cornea. The underlying and surrounding cornea demonstrated stromal infiltrate with a hazy appearance and poorly demarcated edges. A piece of the pigmented plaque was excised at the slit lamp and was sent for culture and histopathology. He was started empirically on moxifloxacin drops hourly for presumed infectious keratitis. Gram stain revealed Gram-positive cocci and cultures grew Streptococcus viridans and Haemophilus species resistant to moxifloxacin, but sensitive to cefazolin. The moxifloxacin was changed to fortified cefazolin 50 mg/ml, as well as gentamicin 14 mg/ml for synergy, and his infectious keratitis improved markedly. At follow-up visits, the superficial pigmented plaque was further debrided, but the intrastromal component accumulated within the margins of his large persistent epithelial defect. Histopathology revealed pigmented acellular material on hematoxylin/ eosin stain sections that stained positively for melanin by

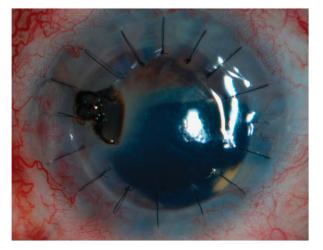


Figure 1 Slit lamp photograph of the pigment deposition in the cornea.