9. Dehghani A, Rezaei L, Tavallali A, Dastborhan Z. Upper eyelid silicone oil migration after sutureless 23-gauge vitrectomy. *Adv Biomed Res*. 2017;6:58.
10. Donker DLT, Paridaens D, Mooy CM, van den Bosch WA. Blepharoptosis and upper eyelid swelling due to lipogranulomatous inflammation caused by silicone oil. *Am J Ophthalmol*. 2005;140:934–6.
11. Osaki TH, Osaki MH, Allemann N, Osaki T. Silicone migration: an unusual eyelid complication following intraocular surgery. *Aesthet Surg J*. 2016;36:20–2.
12. Quintyn JC, Genevois O, Ranty ML, Retout A. Silicone oil migration in the eyelid after vitrectomy for retinal detachment. *Am J Ophthalmol*. 2003;136:540–2.

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Canalicular melt secondary to *Pseudomonas aeruginosa* infection in a pediatric patient

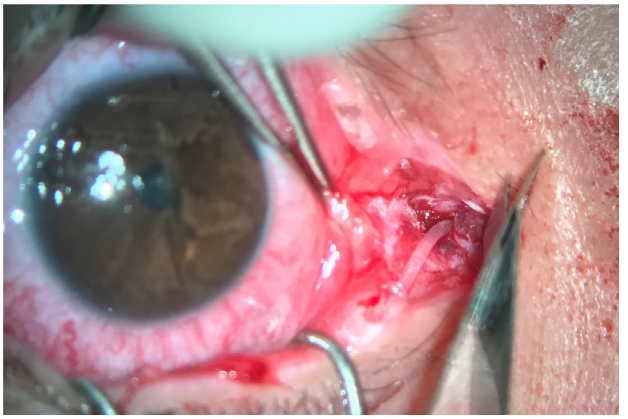
Pseudomonas aeruginosa can cause ocular or periocular infections, such as keratitis, conjunctivitis, and dacryocystitis.[1] There have been reports of periocular and eyelid necrotizing fasciitis secondary to *Pseudomonas* infection and these cases are usually accompanied by systemic illness, malnutrition, alcoholism, or minor ocular trauma.[2] Typically, bilateral ocular involvement and neutropenia is seen. This is the first documented case of unilateral, localized canalicular melting caused by *Pseudomonas* microbial infection.

Case Report

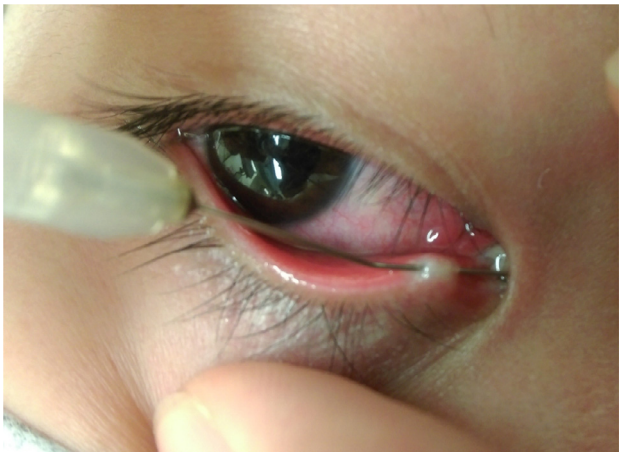
A 14-year-old girl presented with fever, swelling, redness, and pain over the right medial eyelid. She complained of epiphora, foreign body sensation, and mild blurred vision without a history of trauma to the right eye. On examination, she had a visual acuity of 20/20 bilaterally without limitations of extraocular movements. Eye examination was remarkable for congested conjunctiva and tenderness with palpation of the medial right eyelid with purulent discharge from the puncta noted.

Initially diagnosed with dacryocystitis with culture positive for *Pseudomonas aeruginosa*, a follow-up examination showed upper and lower canalicular melting of the right eye (Fig. 1A, 1B). In her hospital course, the patient developed prolonged neutropenic fever. On physical exam, she developed malar rash, oral mucosal changes, and joint stiffness. Labs revealed low complement levels, high ANA (antinuclear antibody), and high ds-DNA leading to a diagnosis of systemic lupus erythematosus (SLE). She was started on high-dose prednisone and hydroxychloroquine, which resolved her neutropenia.

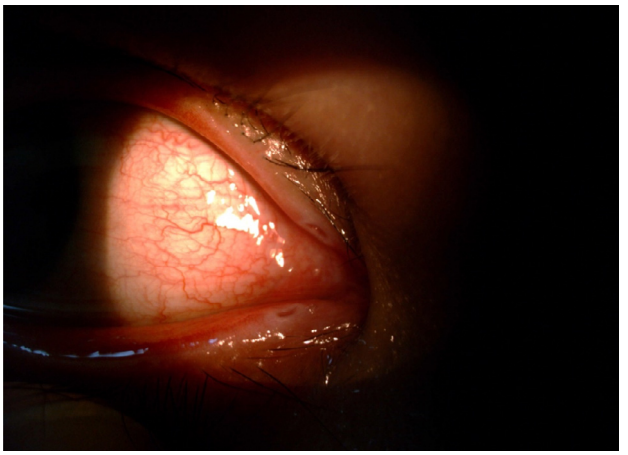
The patient was treated with cefepime and piperacillin and was taken to the operating room for debridement, repair, and silicone intubation of the upper and lower canaliculi (Fig. 1C). Three months after the operation, the silicone tubes were removed. The upper and lower canalicular structures were



Panel C. Intraoperative silicone intubation of the upper and lower canaliculi of the right eye.



Panel A. Right upper eyelid with probed canalicular disruption and associated purulent drainage and chemosis.



Panel D. Post-operative photo of reconstructed and healed right eyelid canalicular system.

Fig. 1—A. right upper eyelid with probed canalicular disruption and associated purulent drainage and chemosis. B, right lower eyelid with probed canalicular disruption and associated purulent drainage and chemosis. C, intraoperative silicone intubation of the upper and lower canaliculi of the right eye. D, post-operative photo of reconstructed and healed right eyelid canalicular system.

normal on exam (Fig. 1D) and the lacrimal drainage system was patent on syringe irrigation test at a 1-year follow-up.

Discussion

Eyelid or periocular necrosis due to infection has been described in several reports, most of which are secondary to trauma such as animal bites or avulsive lacerations.[1-7] Risks increase in those with systemic diseases such as diabetes, alcoholism, or any immunocompromised condition.[7] The most common etiology of periorbital necrotizing fasciitis is *Streptococcus pyogenes* or *Staphylococcus aureus*, but rarely *Pseudomonas aeruginosa* have been identified.[1-7] Non-infectious lid destruction has been described from the bite of the brown recluse spider[4,8] or neoplastic processes like sebaceous gland carcinoma.[4]

Tissue destruction such as necrotizing fasciitis is uncommon in the eyelids due to a rich blood supply; that being said, it can still be fatal.[6] *Pseudomonas aeruginosa* attaches to sialic acid receptors by attaching to the fimbriae; the bacteria then proliferates and releases enzymes such as elastase, alkaline protease, and exotoxin A, *resulting in tissue necrosis*.[3]

In a study of canalicular laceration, 55% of cases were caused by direct penetrating injury. Injury to the lacrimal duct system can lead to scarring, stenosis, and epiphora if not repaired in a timely fashion. In traumatic disruption of the canaliculus, it is common to repair using silicone tubing as a lacrimal stent.[9,10]

In this case, the patient was neutropenic secondary to an SLE flare. Most cases of *Pseudomonas*-related eyelid necrosis have shown association with neutropenia secondary to systemic conditions such as septicemia, malnutrition, cancer, and other types of immunocompromised conditions. There has not been a documented case of such localized canalicular injury as seen in our patient.

Vasculitis has been documented in up to 36% of SLE patients; of those patients, cutaneous manifestations (petechiae, palpable purpura, cutaneous infarction, superficial ulceration, etc.) were the most common and indicate small vessel involvement.[11] The inflammation of these vessels can lead to ischemia of downstream tissue via antibody complex and complement aggregation and deposition. This ischemia combined with low complement levels predisposes the area to infection—in this case, likely predisposing the patient to the localized *Pseudomonas* infection and destruction of the pericanalicular structures. That being said, typically cutaneous presentations of vasculitis in SLE patients are much more widespread and not so localized.

Early diagnosis of the patient's underlying condition was important in this case as resolution of neutropenia has been associated with improved clinical improvement. We were fortunate to isolate the *Pseudomonas* culture early, which may have prevented progression from tissue and muscle necrosis to true necrotizing fasciitis and also allowed for timely repair of the canalicular disruption. If left untreated, the patient may have had devastating, irreparable necrosis and destruction of the lacrimal drainage system.

Conclusions

Although it is rare, it is important to rule out tissue necrosis secondary to *Pseudomonas* in periocular infection. Early diagnosis and recognition of a systemic medical illness in the setting of eyelid necrosis caused by *Pseudomonas aeruginosa* is an important step to prevent possible progression to necrotizing fasciitis and key to a good clinical outcome after treatment and therapy. Thorough evaluation of the eyelid and examining for canalicular disruption helps with initiation of repair as quickly as possible to prevent scarring and stenosis of the lacrimal drainage system.

Supplementary materials

Supplementary material associated with this article can be found in the online version at doi:10.1016/j.jcjo.2018.07.009.

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References

1. Lattman J, Massry GG, Hornblass A. Pseudomonal eyelid necrosis: clinical characteristics and review of the literature. *Ophthal Plast Reconstr Surg*. 1998;14:290–4.
2. Comaish I, Thaller V, Newman P. Necrosis of the lid due to *Pseudomonas aeruginosa*. *Eye*. 2000;14:387–9.
3. Kreger AS. Pathogenesis of *Pseudomonas aeruginosa* ocular diseases. *Rev Infect Dis*. 1983;5:S931–5.
4. Kronish JW, McLeish WM. Eyelid necrosis and periorbital necrotizing fasciitis. *Ophthalmology*. 1991;98:92–8.
5. Lazzeri D, Lazzeri S, Figus M, et al. Periorbital necrotising fasciitis. *Br J Ophthalmol*. 2010;94:1577–85.
6. Rodriguez-Gonzalez F, Marrero-Saavedra D, Rutllan-Civit J, Cabrera-Vargas E, Martinez-Qintana E. Ocular necrotizing fasciitis due to pseudomonas aeruginosa in a non-neutropenic patient. *Saudi J Ophthalmol*. 2013;27:281–2.
7. Prendiville KJ, Bath PE. Lateral cantholysis and eyelid necrosis secondary to pseudomonas aeruginosa. *Ann Ophthalmol*. 1988;20:193–5.
8. Zeligowski AA, Peled U, Wexier MR. Eyelid necrosis after spider bites. *Am J Ophthalmol*. 1986;101:254–5.
9. Jordan DR, Ziai S, Gilberg SM, Mawn LA. Pathogenesis of canalicular laceration. *Ophthal Plast Reconstr Surg*. 2008;24:394–8.
10. Reifler DM. Management of canalicular laceration. *Surv Ophthalmol*. 1991;36:113–32.
11. Kenneth TC, Balabanova M. Vasculitis in systemic lupus erythematosus. *Clin Dermatol*. 2004;22:148–56.

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Intravenous rituximab for the treatment of relapsing adult-onset asthma with periocular xanthogranuloma

In 1993, Jakobiec et al. first reported adult-onset asthma with periocular xanthogranuloma (AOAPOX) in 6 patients, who were treated with oral and inhaled steroids.[1] Clinically, these patients present to the ophthalmologist with bilateral, yellowish-orange, elevated, and indurated xanthomatous eyelid mass lesions.[1] The anterior orbital fat, extraocular muscles, lacrimal gland, and optic nerve may eventually get involved. The onset of asthma may occur concurrently, precede, or follow the periocular/orbital lesions.[2,3] Hence, a timely systemic workup is crucial to identify coexisting severe morbidities like asthma, paraproteinemias, and lymphomas.[2]

The histopathology and immunohistochemistry features eosinophilic infiltrates with dendritic cells, xanthoma cells, lymphocytes, and high CD8 and CD20 positivity.[2–5] A magnetic resonance imaging (MRI) of the orbits is preferred over a computed tomography before planning an incisional biopsy, as MRI provides better soft-tissue delineation, picks up subtle infiltrations and helps in differentiating orbital inflammatory disorders.[6]

The primary treatment for AOAPOX is medical. The corticosteroids, methotrexate, and rituximab have been tried as monotherapies or in combination therapies with satisfactory success.[1,2,7,8] Recently, rituximab monotherapy successfully treated bilateral orbital necrobiotic xanthogranuloma with CD20 predominance.[9] Pomponio et al. used rituximab as first-line monotherapy in AOAPOX associated with IgG4-related disease and reported a sustained (1 year) clinical response after its single course of 2 g in 2 doses.[10] We report 2 patients of AOAPOX who relapsed after stopping oral deflazacort and showed long-term remission after intravenous rituximab therapy.

Case descriptions

Case 1

A 43-year-old male complained of painless, slowly progressive swellings of both upper eyelids of 2 years' duration. He was a known case of bronchial asthma and was currently on inhalational steroids. He was referred to us with a diagnosis of bilateral orbital lymphoma. On examination, the best corrected visual acuity was 6/6 in both eyes with normal intraocular pressures. Both superior orbital sulci were full with medial and central prominences (Fig. 1A–C). He had bilateral upper blepharoptosis with restricted elevation (Fig. 1D) and no proptosis. The overlying eyelid skin appeared healthy and was freely mobile. On palpation, the superior orbital mass lesions were indurated, nontender, and measured 40 × 25 mm. The anterior and posterior segment was within normal limits (WNL).

The working diagnosis of orbital lymphoma and orbital xanthogranuloma was kept. The laboratory investigations were requested, and the complete blood counts, peripheral blood film, lipid profile, and renal and liver function tests were WNL. MRI orbits showed bilateral, ill-defined, hyperintense mass with diffuse soft-tissue infiltration in superior orbits, on both T1W and T2W images (Fig. 1E,F). Bilaterally, the lacrimal glands appeared to be involved, and the extraocular muscles were enlarged. A bilateral incisional biopsy from the superior orbit was performed and detailed histopathology with immunostaining was requested. The biopsy results showed dense nodular collections of xanthoma cells in perivascular and periadenexal locations. The foamy histiocytes were highlighted by CD68 immunostain, which was consistent with features of a diffuse xanthomatous lesion.

Oral deflazacort was started at a dosage of 30 mg/day, 18 mg/day, and 6 mg/day for the first, second, and third month, respectively, with a satisfactory response. After 3 months of stopping steroids, he had an ophthalmic and pulmonary relapse. Intravenous rituximab (375 mg/m^2) was prescribed weekly for 4 weeks followed by a monthly dose for 3 months. At 14 months follow-up after cessation of rituximab (Fig. 2),