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CASE REPORT

Multi-viral canaliculitis: case report and review of literature

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Abstract
Aim To report an atypical case of multiple viruses causing canaliculitis.
Methods Case report of a young female presenting with atypical course of refractory unilateral canaliculitis with complete mid-bicanalicular obstructions. Canalicular scrapings were subjected to immunofluorescence techniques and polymerase chain reactions to identify the viruses.
Results Investigations revealed a canaliculitis of multi-viral etiology; herpes simplex virus and varicella zoster virus. A canalicular curettage followed by topical acyclovir helped in the resolution of canaliculitis.
Conclusion An encounter with an atypical canaliculitis with negative bacteriology work up, suboptimal response to routine therapies, and mid-canalicular obstructions should alert the physician to investigate for viral etiology.

Keywords Viral · Herpes simplex · VZV · Canaliculitis

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Introduction

Viral infections of the lacrimal drainage system are not commonly reported. Lacrimal drainage system is involved in infections with herpes simplex virus (HSV), varicella zoster virus (VZV), trachoma, and infectious mononucleosis [1–7]. HSV and VZV infections account for many cases of acquired canalicular obstructions [1–4]. Most patients are young with a female predilection, present unilaterally, and invariably involve both the upper and lower canaliculi at the mid canalicular level [1–11]. We present a young female with unilateral canaliculitis refractory to conventional treatment. Canalicular scrapings and subsequent real-time polymerase chain reaction (PCR) revealed an infection with a combination of HSV and VZV. To the best of authors' knowledge, this is the first case report of combined HSV and VZV canaliculitis.

Materials and methods

A female of 22 years presented with swelling of the inner sides of left eyelids of 2 months duration (Fig. 1a). The patient was asymptomatic before. The swelling was associated with constant epiphora and discharge. The punctal and canalicular region was inflamed with exuding discharge (Fig. 1b). The right eye showed upper and lower single-wall canalicular aplasia and was asymptomatic. An impression of left

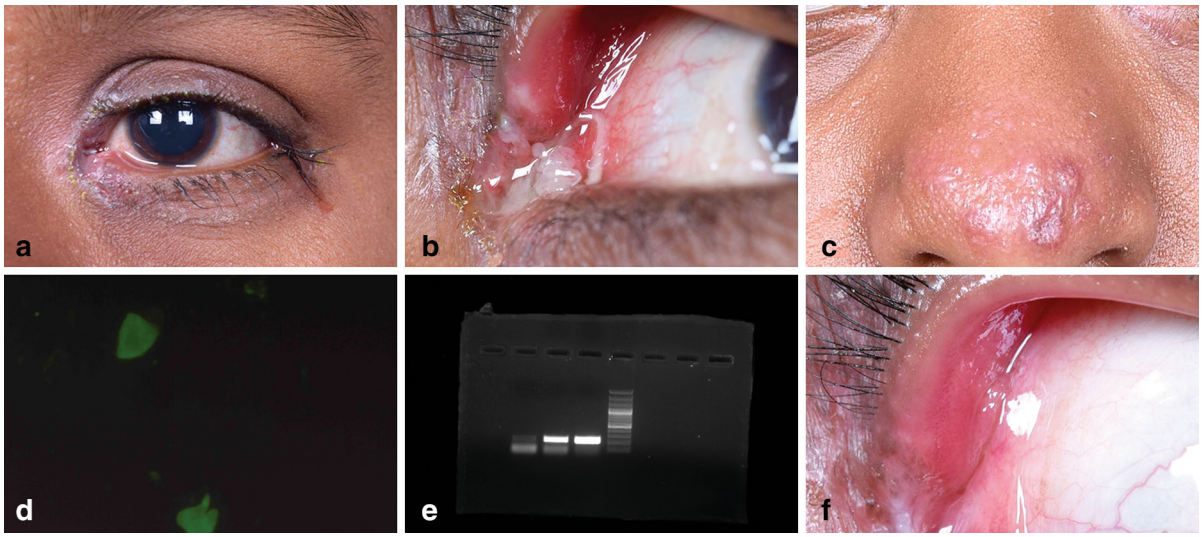

Fig. 1 Clinical photograph showing left upper and lower eyelid swelling over the canalicular portion. Also note the few vesicles on the upper lid (**a**). Inflamed canaliculi with copious discharge in the vicinity (**b**). Healed skin lesions on the tip of the nose (**c**). Immunofluorescence assay showing presence of HSV-1 antigen in epithelial cells (×400) (**d**). PCR showing the presence of HSV-1 DNA (glycoprotein D gene) at 221 base pair region (**e**). Clinical photograph showing a good response in just 2 weeks with topical acyclovir ointment (**f**)

canaliculitis was made, and the patient underwent punctal dilatation and expression of canalicular contents. During expression of the contents, a bicanalicular obstruction (4 mm from upper punctum and 3 mm from lower punctum) was noted. Microbiological workup failed to reveal any organism. The patient returned to clinic with recurrence of symptoms, and microbiological workup of the contents expressed the second time showed *Staphylococcus aureus*. The patient was started on topical antibiotics based on the culture sensitivity profile; however, the response was suboptimal. Detailed examination revealed mild granulation tissue at the edges of both the left upper and lower punctum and proximal vertical canaliculi. The clinical scenario was revisited in view of the atypical course. Left upper lid and the nasal tip had healed vesicular lesions with mild intervening scarring (Fig. 1a, c). The patient denied any past history of blepharoconjunctivitis or rash and was not on any antiviral medications in the past. Corneal evaluation did not reveal any signs of a past herpes infection. The patient underwent punctal dilatation with scrapings of the granulation tissue from the vertical canaliculi. In view of the atypical clinical course, suboptimal response to antibiotics, bicanalicular obstruction, and signs of papular rash, an impression of a possible viral canaliculitis was made. The discharge and tissue scrapings were sent for microbiological evaluation with a specific request for HSV, VZV PCR. In addition, the tissue scrapings were sent for histopathology.

Results

Immunofluorescence assay (IFA) was performed using rabbit anti-HSV-1 polyclonal antibody (DAKO, Denmark) and showed a positive result (Fig. 1d). Conventional PCR techniques were used initially to detect HSV-1 and were found to be positive (Fig. 1e). The presence of HSV-1 and HSV-2 and/or VZV by real-time PCR (R-gene® kit, Argene/BioMérieux) according to the manufacturer's instructions on the Applied Biosystems Real Time 7900 instrument (ABI7900HT, Applied Biosystems, CA, USA) was then performed, and this also showed a positive result for both HSV-1 and VZV. Culture did not grow any bacteria.

Histopathology revealed features consistent with a nonspecific granulation tissue. The patient was diagnosed as multi-viral canaliculitis and was started on topical acyclovir five times a day. The patient showed a good response to the treatment (Fig. 1f) in 2 weeks with complete resolution of discharge and gross

reduction in inflammation. The patient was advised for continuing topical acyclovir for 6 weeks, and the option of conjunctivodacryocystorhinostomy after complete resolution of canaliculitis has been discussed.

Discussion

Lacrimal drainage obstructions have been reported in cases of ocular vaccinia, chicken pox, herpes simplex infections, and herpes zoster ophthalmicus [1–11]. Unlike bacterial and mycotic canaliculitis, viral canaliculitis usually results in permanent and recalcitrant canalicular obstructions. This can be explained by the fact that bacterial infections affect the subepithelial tissues while sparing the epithelial layers and hence the canalicular patency is restored following resolution of infection and inflammation. However, in viral canaliculitis, the epithelial layers are involved, which leads to necrosis of the lining epithelium and replacement by fibrous tissues [5, 6].

Bouzas [2] in 1965 reported two cases of canaliculitis, associated with concurrent infection of eyes with herpes simplex and herpes zoster ophthalmicus, respectively. Subsequently, the same author in 1973 [1] reported 4/19 patients with herpes zoster ophthalmicus and 3/29 patients with herpes simplex to have punctal stenosis and/or canalicular obstruction. Coster and Welham [4] in 1979 reported 20 cases of canalicular obstruction secondary to herpes simplex infection. All the patients were in the first three decades of their lives, and 17 out of 20 patients were females. All patients had mid-zone (2–6 mm from the punctum) canalicular obstruction. Most patients had history of viral conjunctivitis and lid vesicles except three patients where virus isolation was positive without any apparent active herpetic process.

De Koning et al. [5] presented six cases of herpetic canalicular obstructions, all were females with unilateral presentation and four had mid-zonal bicanalicular obstructions. Two of these patients were treated with conjunctivodacryocystorhinostomy with Lester-Jones tubes. Harris et al. presented five female patients (second decade of life) of unilateral herpetic canalicular obstructions, of which three were mid-zonal in location. The current case also fits into the usual profiles in literature (young, female, unilateral, mid-zonal); however, there was no preceding history of a herpes infection, no signs of keratitis and isolation of both HSV and VZV viruses.

In summary, we describe an additional new finding of multiple virus isolation in a case of canaliculitis. An encounter with an atypical canaliculitis with negative bacteriological workup, suboptimal response, and canalicular obstruction should alert the physician to investigate for viral causes.

Compliance with ethical standards

Conflict of interest The authors declare that they have no competing interests.

Disclosure Dr. Ali receives royalties from Springer for his textbook "Principles and Practice of Lacrimal Surgery" and also for his treatise 'Atlas of Lacrimal Drainage Disorders'. Other authors have none.

Patient consent The patient/next of kin/guardian has consented to the submission of the case report for submission to the journal.

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