

Case report

Canalicular pyocele: A new entity and literature review



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Abstract

Canaliculocelles are rare, and under-recognized lesions of the upper lacrimal drainage system that may present as medial canthal masses or swelling. The current case is that of an 85-year-old female patient presented with a painful right upper lid medial, cystic swelling of 10-day duration involving the punctum and the upper canaliculus. A literature review showed that this is the seventh reported case of a canaliculocelle but the first case of acute onset that is associated with inflammatory signs and symptoms and accumulation of purulent material. The term canalicular pyocele is suggested to describe this condition.

Keywords: Canaliculocelle, Canaliculops, Lid lesion

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Introduction

Canaliculocelle or canaliculops, first described by Fouad AR in 1972¹ and studied by Sacks et al. in 1987,² is a rare form of noninflammatory and noninfectious canalicular distension that may present as a medial eyelid cystic swelling.^{3,4} Although rare, the differential diagnosis of cystic lesions in the medial canthus area should include canaliculocelles.³ Recently, Javed et al.⁴ and Yoon et al.⁵ described the detailed histopathological and immunohistochemical characteristics of a canaliculops and its differentiation from a simple conjunctival cyst. The current report is the seventh case of a canaliculocelle, but it is the first report to show an association with signs and symptoms of inflammation with acute onset.

Case report

An 85-year-old woman, diabetic and hypertensive for the previous 12 years, presented with a progressive, painful, red, cystic swelling of 10-days duration over the right upper punctum and the canaliculus (Fig. 1A). She denied a history of discharge, epiphora, conjunctival injection, or irritation, trachoma, herpetic keratitis, previous lid surgery, or trauma. The ophthalmic history revealed right quiet pseudophakia

from 5 years previously. Examination revealed an approximately 8 × 8-mm opaque, immobile, erythematous, cystic swelling of the right upper medial eyelid that was centered on the punctal papilla extending to the medial corneal limbus and the right lower punctum (Fig. 1B). Lid eversion was impossible because of lesion tenderness. Acute, suppurative canaliculitis was suspected initially, but there was no discharge upon pressure, regurgitation, or pouting of the punctum. In fact, the punctal orifice was occluded and barely visible. The left eye had a central corneal opacity with poorer vision (counting fingers closely compared to 20/200 in the right eye), but no similar complaints. The patient underwent surgery under local anesthesia and sterile conditions. The punctal orifice was identified, and a membrane occluding the punctal orifice was punctured using a blunt-tipped punctal dilator, which released a large amount of purulent discharge from the lesion (Fig. 2). The upper drainage system was compressed to allow drainage of all purulent materials. Probing of the upper drainage system showed a dilated canaliculus with wide side-to-side movements and a soft stop at the most medial end of the canaliculus. No canaliculiths or sulfur granules were encountered. Irrigation of the lower drainage system was patent into the nose with no reflux. The upper punctum and canaliculus were marsupialized and

Received 24 June 2016; received in revised form 28 August 2016; accepted 6 December 2016; available online 18 December 2016.

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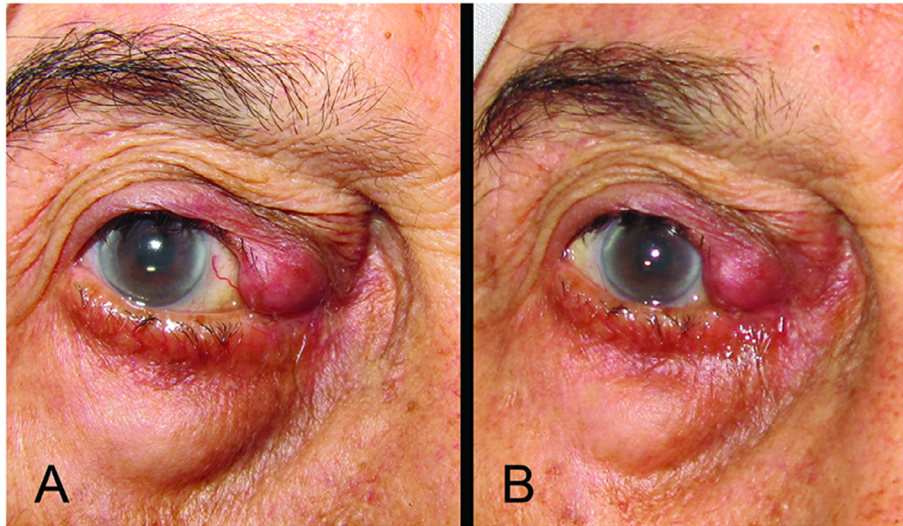


Figure 1. (A) Swelling of the medial upper lid and (B) reaching the medial corneal limbus and the lower lid punctum.

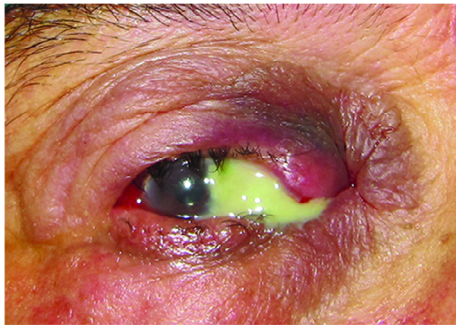


Figure 2. The release of purulent discharge after opening the punctum and before marsupialization.

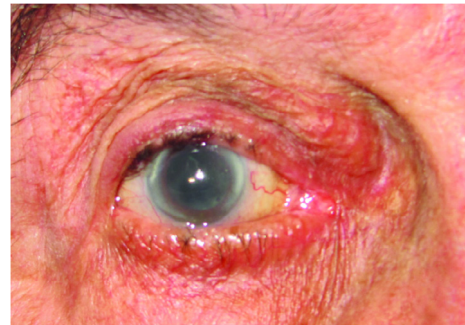


Figure 3. The right upper canaliculus 5 months after marsupialization with no recurrence.

irrigated with 10% Povidone-Iodine solution (Al-Rahma Pharmaceutical Co., Amman-Jordan). The patient was discharged and instructed to apply topical chloramphenicol ointment (Amman Pharmaceutical Industries Co., Amman-Jordan) for four times daily and to take oral Augmentin (Amoclan[®], Hikma Pharmaceuticals, Amman-Jordan) for three times daily. A culture swap of the opened upper canaliculus on the first postoperative visit showed a mixed infection caused by *Klebsiella oxytoca* species that is sensitive to Tigecycline, Cefoxitine, Amicacin and Imipinem, and *Staphylococcus* species coagulase negative that is sensitive to Ampicillin, Chloramphenicol, Clindamycin, Tigecycline and Vancomycin. The pain resolved, with no recurrence nor tearing after more than 8 months of follow-up (see Fig. 3).

Discussion

Canaliculocelles are a rare, under reported and under-recognized lid lesions.^{3,5} It was also referred to as canaliculops in some reports.^{2,4} Sacks et al.² first reported the pathology in 1987. For canaliculops to form, both the medial and lateral ends of the canaliculus must be obstructed.^{2,5} The common features of all the convincing reported cases of canaliculops are their clinical chronicity (range, 2 months to 20 years) with a median 1-year duration, no tenderness on palpation, and the absence of overt inflammatory signs.^{3,5}

A slight female preponderance was noted (5/7) among all patients, including the current patient.

Whereas six cases of canaliculocelles (canaliculops) have been reported in the literature,²⁻⁵ none was painful or showed signs of inflammation or tenderness on palpation (Table 1). The current case is the first reported case of an inflamed and painful canaliculocelle with purulent collection, referred to as a canalicular pyocele.

Similar to previous cases,^{2,4,5} the current patient had no history of epiphora, and there was a soft stop at the medial extent of the dilated canaliculus. Previous reports also have reported punctal stenosis secondary to punctal cautery for dry eyes⁵ or punctal agenesis.⁴ However, the current patient had no previous lid trauma, punctal cautery, or surgery but had an effaced upper punctum on slit-lamp examination.

The differential diagnosis of canaliculocelle includes medial chalazion, conjunctival cysts, canalicular diverticulum in non-inflammatory cases, and canaliculitis (primary or secondary) if inflammation was present.^{2,5} Although this case had inflammatory signs, there was no history of epiphora, recurrent conjunctivitis, ocular surface discomfort, eyelid trauma, punctal surgery, or plug insertion differentiating it from suppurative canaliculitis.⁶ Furthermore, canaliculitis usually has a longer duration of symptoms,⁷ and canalicular diverticulum presents with epiphora in a patent nasolacrimal system, while

Table 1. Literature review of canaliculocelles and their findings.

Author	Number of patients	Age (years)/sex	Upper / lower canaliculus	Duration of symptoms	Treatment	Cultures/pathology	System patency
Sacks et al. ²	1 case	40 male	Right upper	1 year	Excisional biopsy	Cystic space lined by hyperplastic multilaminar nonkeratinizing squamous epithelium	N/A
Kim et al. ³	2 cases	77 male	Right medial canthus	1 year	Excisional Biopsy with canaliculo-dacryocystorhinostomy with silicone intubation	Chronic inflammation, no foreign bodies, and a cyst lined by nonkeratinized stratified squamous epithelium.	Upper canalicular obstruction
		40 female	Left lower	20 years	Transconjunctival surgical excision	nonkeratinized stratified squamous epithelial lining without inflammatory cells or foreign bodies.	Occluded left lower punctum
Javed et al. ⁴	1 case	65 female	Right upper	1 year	Excisional biopsy.	Nonkeratinized stratified squamous epithelial, Positive CK 7, 14, 17	Soft stop medially
Yoon et al. ⁵	2 cases	66 female	Right lower	2 months	Incisional biopsy with posterior marsupialization	Positive CK 7, 14, 17, 18	Soft stop medially
		53 female	Right upper	2 years	Incisional biopsy with posterior marsupialization	Positive CK 7, 14, 17, 18	Soft stop medially
Ababneh	1 case	85 female	Right upper	10 days	Incisional biopsy with posterior marsupialization	Klebsiella oxytoca species and Staphylococcus species coagulase negative	Soft stop medially

canaliculops had punctal and/or canalicular obstruction and present as a cystic lesion.⁸

The previous reports^{2,3,5} showed nonpurulent and nonviscous watery secretions from the lumen of the canaliculocelle cases that can be attributed to the fact that there are no keratinous components or accessory mucous glands in the wall of the normal canaliculus.⁹ However, purulent material was released once the punctal orifice was opened, supporting the presence of an inflammatory process.

Javed et al.⁴ and Yoon et al.⁵ described the detailed histopathological and immuno-histochemical characteristics of a canaliculops. The cystic wall is lined with multilayered nonkeratinizing stratified squamous epithelium. The epithelium is packed tightly with orderly arranged cells, and the characteristic regimented basal layer is arranged in a palisading fashion, with no goblet or subepithelial inflammatory cells, thus differentiating it from conjunctival cysts.^{4,5}

The treatment described previously consisted of surgical excision^{3,4} or marsupialization through the conjunctiva.^{2,5} In the current case, marsupialization was done to preserve the lacrimal drainage passages followed by irrigation with 10% Povidone-Iodine solution, and topical and systemic antibiotics resulted in improved symptoms and no recurrence during the follow-up period.

Conflict of interest

The author declared that there is no conflict of interest.

References

1. Fouad AR. A rare case of canaliculocelle. *Bull Ophthalmol Soc Egypt* 1972;**65**:265–7.
2. Sacks E, Jakobiec FA, Dodick J. Canaliculops. *Ophthalmology* 1987;**94**:78–81.
3. Kim JC, Ko YH, Woo KI, Kim YD. Canaliculocelle presenting as a medial canthal mass. *Ophthal Plast Reconstr Surg* 2009;**25**:236–8.
4. Javed Ali M, Saha D, Kumar Mishra D, Naik MN. Canaliculops associated with punctal agenesis: a clinicopathological correlation and review of literature. *Ophthal Plast Reconstr Surg* 2015;**31**: e108–11. <http://dx.doi.org/10.1097/IOP.0000000000000120>.
5. Yoon MK, Jakobiec FA, Mendoza PR. Canaliculops: clinicopathologic features and treatment with marsupialization. *Am J Ophthalmol* 2013;**156**, 1062–1068.e1. <http://dx.doi.org/10.1016/j.ajo.2013.06.008>. Epub 2013 Aug 12.
6. Vecsei VP, Huber-Spitzy V, Arockar-Mettinger E, Steinkogler FJ. Canaliculitis: Difficulties in diagnosis, differential diagnosis and comparison between conservative and surgical treatment. *Ophthalmologica* 1994;**208**:314–7.
7. Zaveri J, Cohen AJ. Lacrimal canaliculitis. *Saudi J Ophthalmol* 2014;**28**:3–5. <http://dx.doi.org/10.1016/j.sjopt.2013.11.003>, Epub 2013 Nov 13.
8. Adjemian A, Burnstine MA. Lacrimal canalicular diverticulum: a cause of epiphora and discharge. *Ophthal Plast Reconstr Surg* 2000;**16**:471–2.
9. Yonekawa Y, Jakobiec FA, Zakka FR, Kim N. Keratinizing cyst of the lacrimal punctum. *Cornea* 2013;**32**:883–5.