Case Series and Brief Reports

Canalicular Cyst

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Key Words

Eyelid · Canalicular cyst · Diverticulum · Canaliculocoele · Dacryops

Abstract

Purpose: To report a new entity discovered in the lacrimal drainage system, a congenital canalicular cyst (as opposed to a diverticulum or canaliculops). **Methods:** Clinical, radiographic, histopathologic and immunohistochemical findings were reviewed. **Results:** A 49-year-old man developed a left upper eyelid cyst seen by CT scanning to be attached to a small bony spicule. No connection to the lacrimal sac or canaliculus was discovered during surgical excision. Histopathologically, the cyst was lined mostly by canalicular epithelium, expressing a cytokeratin profile similar to that of a normal canaliculus and canaliculops. **Conclusion:** This case increases ophthalmologists' appreciation of the range of conditions that can affect the lacrimal drainage and canalicular system. The treatment for a diverticulum is marsupialization, whereas that for a canalicular cyst is complete excision.

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Introduction

The canaliculus has been documented to be a site for a variety of pathologic conditions including squamous papillomas, ecstasias of the canaliculus (canaliculops) and diverticula [1–6]. We describe a unique, isolated cyst lined by canalicular squamous epithelium.

Case Report

A 49-year-old man slowly developed an uninflamed fluctuant mass in his left upper eyelid without pain, redness, discharge or tearing (fig. 1a). Probing and irrigation demonstrated canalicular patency to the nose. CT disclosed a cystic mass located in the left anterior superior orbit (fig. 1b, top and bottom panels) attached to a small bony spicule. During surgery, a thin-walled cyst was visualized without punctal reflux of fluid during various manipulations. No connection was discovered between the cyst and the lacrimal sac or canaliculus.

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Fig. 1. Canalicular cyst. **a** Mass in the left upper eyelid above the medial canthal tendon. **b** A small bone density (top panel, crossed arrow) was detected at the posterior-medial edge of the lesion. The lesion (top panel, arrow) had a uniform density. In the bottom panel, the well-defined borders of the lesion (arrows) and the projecting bony spur (crossed arrow) can be seen. **c** The cyst was lined by a multilaminar nonkeratinizing squamous epithelium. **d** Partial cystic lining by a double layer of cuboidal epithelium with a prominent fibrous wall (arrows), also shown in the **inset**. **e** The lining displayed a thickness of 12 cell layers. The adluminal cells had protrusions (arrows). The basal germinal cells were regimented (crossed arrows). **f** Abrupt (arrow) or gradual (crossed arrow) transitions of multilaminar canalicular epithelium (CA) into conjunctiva-type epithelium (CO). **c–f** HE. **c** $\times 25$. **d** $\times 10$. **Inset** $\times 100$. **e** $\times 200$. **f** $\times 200$.

Fig. 2. Special stains of the canalicular cyst. **a** Alcian blue stains goblet cells (colors refer to the online version only). **b** CK7 immunostains the superficial layer of the cells below (arrows) and the two layers of conjunctival epithelium above (crossed arrows). **c** CK14 immunostains either the basal and parabasal cells (top panel) or the full-thickness epithelium (bottom panel). **d** CK17 haphazardly immunostains cells throughout all levels of the epithelium (top panel). CK18 immunostains only the most superficial cells (middle panel, arrows). Ki-67 stains a paucity of the germinal basal cells (bottom panel). **a** Alcian blue. $\times 400$. **b–d** Immunoperoxidase reaction, diaminobenzidine chromogen, hematoxylin counterstain. **b** $\times 100$. **c** $\times 200$ (top and bottom panels). **d** $\times 200$ (top and middle panels); $\times 100$ (bottom panel).

Microscopic examination disclosed a cyst lined mostly by a multilaminar (10–12 cell layers thick), nonkeratinizing squamous epithelium with well-regimented basal cells (fig. 1c). Some segments were lined by a double layer of low cuboidal cells (fig. 1d–f). Goblet cells were occasionally detected at the juncture of the two kinds of epithelium (fig. 2a). Immunohistochemical staining for cytokeratins (CK) 7, 14 and 18 disclosed a profile similar to that of the canaliculus and canaliculops (fig. 2b–d, top and middle panels). The basal germinal cells rarely stained positive for Ki-67 (fig. 2d, bottom panel).

Discussion

In contrast to a totally isolated cyst, canaliculops is a distention – and a diverticulum is a saccular outpouching with an orifice – of a segment of the canaliculus [3–6]. In cases of a diverticulum, the nasal aspect of the eyelid is swollen and the punctum is reddened. Persistent tearing and, most importantly, an associated purulent punctal discharge may be encountered. The latter can be provoked by applying pressure over the eyelid swelling. In our case, irrigation through the lower punctum demonstrated partial patency of the drainage system into

the nose, with reflux of the fluid through the upper eyelid punctum. This maneuver may also revealingly produce an additional swelling of the eyelid. A dacryocystogram is the gold standard for establishing the diagnosis, but this procedure is currently rarely performed. Treatment consists of surgical marsupialization of the diverticulum onto the conjunctival fornix [6] or excision and closure with silicone intubation and possible dacryocystorhinostomy if near the common canaliculus.

The lesion described herein transilluminated and was totally separate from the normal lacrimal drainage system, as established clinically by the absence of discharge, the uncompromised canalicular patency during irrigation, the absence of an enlargement of the mass during irrigation and the absence of punctal fluid reflux with overlying digital pressure. Its location above the medial canthal ligament would be exceptional for a canalicular diverticulum [6], which creates a medial eyelid swelling inferior to the medial canthal tendon. A CT study demonstrated an isolated lesion attached to a bony spur of the medial orbital wall near the trochlea. Furthermore, during surgery no orificial connection to the nasolacrimal drainage system was observed. Histopathologic examination disclosed a multilaminar, nonkeratinizing squamous epithelial lining up to 12 cells in thickness and resting on a highly regimented, picket fence-like arrangement of smaller basal germinal cells – the architecture of the normal canalicular epithelium. There were, however, interposed segments of low cuboidal epithelium reminiscent of conjunctiva, suggesting a malformation. The management of a cyst like the present one is complete surgical excision.

The immunohistochemical CK profile was similar to that of the normal canaliculus and canaliculops (CK7 positive in superficial cells, CK14 positive in basal and suprabasal cells, CK17 patchily positive throughout the epithelium and CK18 positive in superficial cells) [4]. It is posited that during the development of the nasolacrimal drainage system, a small sequestration of cells destined to be part of the canalicular epithelium was detached from the developing canaliculus and slowly enlarged over the course of the patient's lifetime to culminate in a clinically relevant cystic mass.

Statement of Ethics

This study was conducted in compliance with the rules and regulations of the Health Insurance Portability and Accountability Act as well as in adherence to the Declaration of Helsinki and all other relevant federal and state laws.

Disclosure Statement

The authors have no financial disclosures or conflicts of interest to report.

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