Canaliculitis in supernumerary puncta and canaliculi

Ku Chui Yong,[1] Tan Aik Kah,[1,2] Faridah Hanom Annuar[1]

[1]Department of Ophthalmology, Faculty of Medicine, University Kebangsaan Malaysia Medical Centre (UKMMC), Cheras; [2]Department of Ophthalmology, Faculty of Medicine and Health Sciences, University Malaysia Sarawak (UNIMAS), Kuching, Malaysia

Abstract

We report the first case of supernumerary puncta and canaliculi presented with canaliculitis. A-59 year-old gentleman presented with painful swelling of the left lower lid for a week, which was associated with epiphora. The swelling was confined to the nasal aspect of the left lower lid (0.5×0.5 mm) with inflamed overlying skin. Two puncta (0.5 mm apart) were noted. The outer punctum at the normal anatomical position was a cul-de-sac while the inner punctum it the caruncle was patent. We described the embryology leading to supernumerary puncta and canaliculi to explain the paradoxical patency of the abnormally located punctum as well as the pathomechanism leading to canaliculitis. The patient was treated with oral cloxacillin 500 mg, 6 hourly for 5 days; the cellulitis subsided after three days.

Introduction

Canaliculitis is a common encounter in ophthalmic practice but supernumerary puncta and canaliculi (SPC) are rare congenital disorders. In a large series, only 23 patients were diagnosed over the period of 20 years.[1] We report a case of canaliculitis which is associated with SPC.

Case Report

A 59-year-old gentleman presented with painful swelling of the left lower lid for a week, which was associated with epiphora. This was his first episode, and he was asymptomatic prior to this presentation. The swelling was confined to the nasal aspect of the left lower lid (0.5×0.5 mm) with inflamed overlying skin (Figure 1A). Eversion of the lower eyelid revealed two puncta, 0.5 mm apart (Figure 1B). The outer punctum was situated at the normal anatomical position; whereas the inner punctum in the caruncle. Gentle pressure did not result in any regurgitation from the both puncta. The patient was treated with oral cloxacillin 500 mg, 6 hourly for 5 days. The cellulitis subsided after three days. Probing and syringing was performed after one week. The inner punctum had a hard stop with patent lacrimal drainage. The outer punctum had a soft stop with regurgitation of fluid from the same punctum. Dacryocystography confirmed the patency of the inner lower punctum. The outer punctum-canaliculus system was a cul-de-sac (Figure 1C). There were no other abnormalities.

Discussion

Most SPCs (78%) present with epiphora. Among the 23 patients reported by Satchi *et al*., none presented with canaliculitis.[1] This is the first report of SPC presented with canaliculitis. Sequestration of tear and debris in the cul-de-sac served as nidus for infection. The resultant canaliculitis with its surrounding edema caused obstruction of the lacrimal drainage; hence epiphora.[2] Epiphora however, may develop despite patent lacrimal drainage system. The 2-compartment model for lacrimal canalicular drainage of Kakizaki *et al.*, suggested that the muscle of Duverney-Horner may deviate normal flow within the accessory canaliculus and thence transport tears back to the lacrimal tear lake, leading to epiphora.[3] The entire lacrimal drainage apparatus is of ectodermal origin. During development, a solid epithelial cord forms in the region of the medial lower eyelid (Figure 2A) and sends projections to form the canaliculi and the nasolacrimal duct (Figure 2B).[4] SPC is due to extra out-budding of the solid epithelial cord (Figure 2C). Canalization begins at 4 months of gestation with disintegration of the central ectodermal core, forming lacrimal drainage outflow system. In this case, the extra inner canalicular epithelial bud (nearer to the main epithelial cord) underwent complete canalization and remained connected to the main epithelial cord. The outer canalicular epithelial bud,

Correspondence: Tan Aik Kah, Ophthalmology Department, Faculty of Medicine and Health Sciences, University Malaysia Sarawak, Lot 77, Sekysen 22 Kuching Town Land District, Jalan Tun Ahmad Zaidi Adruce, 93150 Kuching, Sarawak, Malaysia.
Tel +6082 416550 - Fax + 6082 422564.
E-mail: portwinestain@hotmail.com; aktan@fmhs.unimas.my

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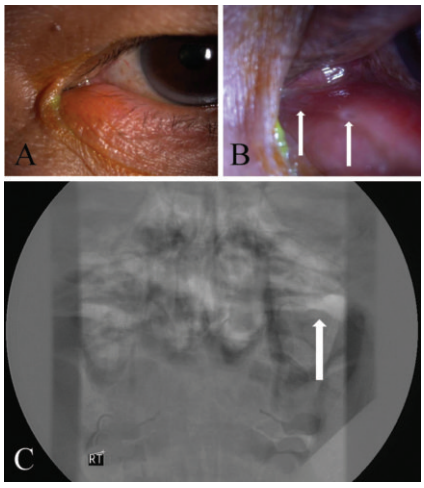


Figure 1. A) Canaliculitis of the left lower eyelid. B) Everted left lower eyelid revealing supernumerary puncta (white arrow). C) Dacryocystography showed pooling of dye in the cul-de-sac (white arrow).

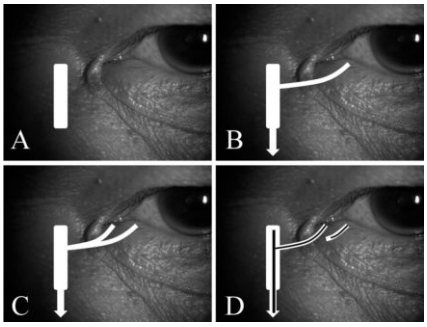


Figure 2. A) Embryology of supernumerary puncta and canaliculi. Formation of a solid epithelial cord in the region of the medial lower eyelid. B) Projections of epithelial cord to form the canaliculi and the nasolacrimal duct. C) Extra out-budding of the solid epithelial cord in supernumerary puncta and canaliculi. D) The outer canalicular epithelial cord was separated from the main epithelial cord, forming a cul-de-sac.

although its punctum is located at the normal anatomical position, was separated from the main epithelial cord; forming a cul-de-sac (Figure 2D).

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