

Actinomyces canaliculitis complicating congenital nasolacrimal duct obstruction in an infant

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Actinomyces israelii is a Gram-positive anaerobic organism commonly associated with canaliculitis in adults. Pediatric canaliculitis is relatively rare, especially in infancy. We report the case of an 11-month-old boy who presented with co-existing canaliculitis and congenital nasolacrimal obstruction. The presenting signs included epiphora, discharge, conjunctival congestion, and matting of lashes. On examination, punctual pouting, regurgitation, and yellow canaliculiths were noted. A punctoplasty and canalicular curettage were performed along with nasolacrimal probing. Microbiological tests confirmed the organisms to be *A. israelii*. We discuss the clinical features and management of *Actinomyces*-associated canaliculitis and review the available literature on pediatric canaliculitis.

Key words: Canaliculith, dacryolith, epiphora, lacrimal duct, punctoplasty, watering

Actinomyces israelii is a Gram-positive anaerobic bacterium that is difficult to isolate and identify. It typically causes infections of hollow spaces such as the canaliculi, with the formation of canaliculiths and is associated with a chronic, purulent, granulomatous infection with the presence of yellowish sulfur granules.^[1] The mainstay of treatment in canaliculitis is punctoplasty and curettage with a low incidence of postsurgical epiphora.^[1-4] Preexisting nasolacrimal duct (NLD) obstruction with canaliculitis is difficult to diagnose, although it has been observed in few adult cases following successful curettage.^[2] There have

been reports of pediatric canaliculitis in literature, but to the best of our knowledge, co-existent congenital nasolacrimal duct obstruction (CNLDO) with canaliculitis has not been previously described in literature.

Case Report

An 11-month-old male child presented with symptoms of watering and discharge in the left eye since birth. The complaints had persisted even after lacrimal sac compression. Redness and discharge in left eye had increased for 3 weeks. In the clinic, the child did not cooperate for an examination with a hand-held slit lamp. A fluorescein dye disappearance test was performed which showed delayed clearance in the left eye. With a diagnosis of the left-sided CNLDO, an examination under anesthesia with irrigation and probing was scheduled.

On examination, the right eye was unremarkable with a patent lacrimal system. The left eye conjunctiva showed some congestion, the cornea was clear and anterior segment, and fundus examinations were normal. Copious discharge was noted over left upper punctum [Fig. 1a]. Using two cotton tip applicators, the canaliculus was squeezed – from the distal part, gradually upward toward the punctum and multiple small yellowish granules were expressed out [Fig. 1b and c]. A vertical incision was made through the posterior wall of the punctum and vertical canaliculus followed by a horizontal incision along a portion of the horizontal canaliculus. All the discharge and granular material were curetted out. The lower punctum was normal, and irrigation through lower canaliculus showed regurgitation through upper punctum suggesting co-existent CNLDO. A hard stop was felt during the irrigation further confirming the location of the obstruction. On probing through lower punctum, a membranous obstruction was encountered at lower nasolacrimal duct opening, which was then overcome. Nasal endoscopy confirmed the presence of probe through NLD opening. Subsequent irrigation was patent. Microbiology of the expressed material showed delicate, branched, Gram-positive filaments irregularly arranged in a background of amorphous material suggestive of *Actinomyces* species [Fig. 2]. The species was subsequently identified as *A. israelii* on the anaerobic blood agar plates. The child was administered fortified cefazolin eye drops for 2 weeks. At 8-month follow-up, the child was asymptomatic.

Discussion

Symptoms of canaliculitis typically include epiphora, chronic conjunctivitis, swelling over the medial canthus, a “pouted” or everted punctum, and purulent discharge.^[2] The presence of

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Table 1: A summary of previously reported cases of pediatric canalculitis

Authors, year	Age/sex	Previous diagnosis	Presenting symptoms	Clinical findings	Organism identified	Treatment	Co-existing conditions	Antibiotic therapy	Outcome	Duration from initial symptom to diagnosis of canalculitis
Pine <i>et al.</i> , 1960 (Case 1) ^[3]	6/male	Conjunctivitis	Discharge	Induration, conjunctival congestion, canalicular concretions, enlarged punctum	<i>A. israelii</i>	Canalicular curettage	Canalicular stenosis	Topical chloramycetin	Resolved	3 years
Pine <i>et al.</i> , 1960 (Case 2) ^[3]	14/male	Conjunctivitis, chalazion	Discharge	Swelling, conjunctival congestion, thickening of medial eyelid	<i>A. israelii</i>	Canaliculotomy and canalicular curettage	None	Neosporin ointment and achromycin ophthalmic oil	Resolved	1 year
Seal <i>et al.</i> , 1981 ^[4]	14/male	Conjunctivitis	Discharge	Swelling, induration	<i>Arachnia propionica*</i>	Canalicular curettage and exploration	Nasolacrimal duct obstruction	Topical and oral penicillin, penicillin syringing	Resolved	2 months
Pavilack and Frueh, 1992 ^[5]	10/female	NA	Epiphora	Punctal regurgitation, pouting punctum, thickening of medial eyelid, concretions	Not reported	Canalicular curettage	None	Topical sulfacetamide sodium	Resolved	20 months
McKellar and Aburn, 1997 ^[1]	10/female	Recurrent conjunctivitis	Discharge	Punctal regurgitation, pouted punctum, erythema of the plica and lower eyelid, concretions	<i>A. israelii</i>	Two-snip punctoplasty, curettage and probing	None	Topical cefazolin	Resolved	6 months
Park <i>et al.</i> , 2004 ^[7]	5/female	Nasolacrimal duct obstruction	Discharge**	Punctal regurgitation, eyelid erythema, concretions	<i>Actinomyces</i> spp., <i>S. viridans</i> , <i>E. corrodens</i> , <i>Peptostreptococcus</i> , <i>Clostridium</i> , and <i>Propionibacterium</i> [§]	Punctoplasty, curettage	Idiopathic thrombocytopenic purpura, past history of immunosuppression	Not specified	Resolved	-
Yuksel <i>et al.</i> , 2012 ^[8]	12/female	NA	Epiphora, discharge	Enlarged punctum, swelling, induration	<i>A. israelii</i>	Canaliculotomy and canaliculoplasty with bicanalicular intubation	None	Topical fortified cefazolin + ciprofloxacin and oral penicillin V	Resolved	7 years
Ding <i>et al.</i> , 2017 ^[9]	6/male	NA	Discharge	Pouting punctum, lacrimal fistula	<i>S. constellatus</i>	Canaliculotomy with fistula excision [†]	Lacrimal fistula	Levofloxacin drops and gatifloxacin ointment	Resolved	5 years

Contd...

Table 1: Contd...

Authors, year	Age/sex	Previous diagnosis	Presenting symptoms	Clinical findings	Organism identified	Treatment	Co-existing conditions	Antibiotic therapy	Outcome	Duration from initial symptom to diagnosis of canaliculitis
Present case	11 months/ female	Congenital nasolacrimal duct obstruction	Epiphora, discharge, recurrent conjunctivitis, epiphora	Punctal regurgitation, pouting punctum, concretions, conjunctival congestion	<i>A. israelii</i>	Canaliculotomy, curettage, antibiotic irrigation, nasolacrimal duct probing	Congenital nasolacrimal duct obstruction	Topical cefazolin	Resolved	2 months

**Arachnia propionica* has been reclassified as *P. propionicus*. **The patient had previously undergone probing with intubation once and balloon dacryoplasty twice: The second time with a monocanaliculostomy placement. †Microscopic examination of the concretions suggested *Actinomyces* spp. Cultures grew *S. viridans*: and moderate *E. corrodens* species. Anaerobic cultures grew *Peptostreptococcus*, *Clostridium*, and *Propionibacterium*. †Initially, the patient underwent a punctum sparing canaliculotomy which was not successful in resolving the symptoms. Brazier and Hall and Kaliki *et al.* in their separate series, have included one child each with canaliculitis: aged 1 month and 96 months, respectively.^[6,11] Similarly, Smith in his series has reported a case of *actinomyces* related canaliculitis in a 17-year-old female.^[10] However, the details of those patients were not specified in their papers and therefore, those cases are not included in this table. NA: Not available, *A. israelii*: *Actinomyces israelii*, *S. viridans*: *Streptococcus viridans*, *E. corrodens*: *Eikenella corrodens*, *S. constellatus*: *Streptococcus constellatus*, *P. propionicus*: *Propionibacterium propionicus*

“yellow sulfur granules” at the punctum is a pathognomonic feature of *Actinomyces* canaliculitis. Pediatric canaliculitis is uncommon, and a review of the available English literature showed only 11 previous cases, the findings of which are tabulated in Table 1.^[1,3-10]

The different treatment options for canaliculitis described in the literature include curettage with punctoplasty (one snip to enlarge the punctum) and canaliculotomy (enlarging the punctum incision along the canaliculus).^[1,12] However, scarring and dysfunction of the lacrimal pump can occur following canaliculotomy; therefore, some authors have also tried canaliculoplasty (narrowing the dilated canaliculus using a 6–0 polyglactin suture) along with lacrimal intubation using Crawford stents.^[8] Our technique of management described in this communication is similar to the one described by Perumal and Meyer. Which consisted of a 2-mm vertical canaliculotomy with sharp-tipped scissors followed by retrograde expression of the canalicular contents by compressing the canaliculus medial to lateral with 2 cotton-tipped applicators.^[13] Conservative treatment in the form of punctal dilatation, canalicular expression, and topical antibiotics has also been tried for treating canaliculitis.^[11] However, conservative treatment alone results in incomplete resolution often necessitating additional procedures such as punctoplasty with canalicular curettage.^[11] In principle, enlarging the punctum and a thorough curettage of all concretions followed by comprehensive antibiotic coverage based on the sensitivity of the cultured organism is essential to treat canaliculitis.

Actinomyces are normal commensal bacteria in humans and primarily cause opportunistic infections during immunosuppressive state or when loss of continuity of epithelial lining in mucosa occurs.^[3,12,14] In our case, there seems to be no certain predisposing factor for *Actinomyces* infection to occur. In our case, it is difficult to establish conclusively if the canalicular infection had any role to play in the development of nasolacrimal duct obstruction (NLDO). It has been postulated that the presence of bacteria in the lacrimal system could initiate an inflammatory response and result in fibrosis and subsequently cause NLDO.^[15] However, the classical history of epiphora soon after birth, the typical membranous obstruction felt during probing which could be easily overcome and visualized endoscopically; and the uneventful recovery after probing suggests that in our case, the NLDO was congenital in origin and the co-existence of canaliculitis was largely fortuitous.

Kaliki *et al.* reported that in canaliculitis, the mean duration of symptoms until diagnosis is 10 months (range = 1 month–5 years) in their cohort, which predominantly included adults.^[11] In addition, while this series had pediatric cases from age 8 upward, details of individual cases were not provided. However, in children, literature suggests that the mean duration of symptoms until a diagnosis is significantly higher at 27.7 months (range = 2 months–7 years) [Table 1]. Furthermore, among the reported cases, the most common condition that the patients were diagnosed as having before definitive diagnosis and treatment for canaliculitis was conjunctivitis (4/6; 66.7%).

Conclusion

Canaliculitis in children is uncommon, and literature suggests that in children, NLDO and chalazion are common conditions

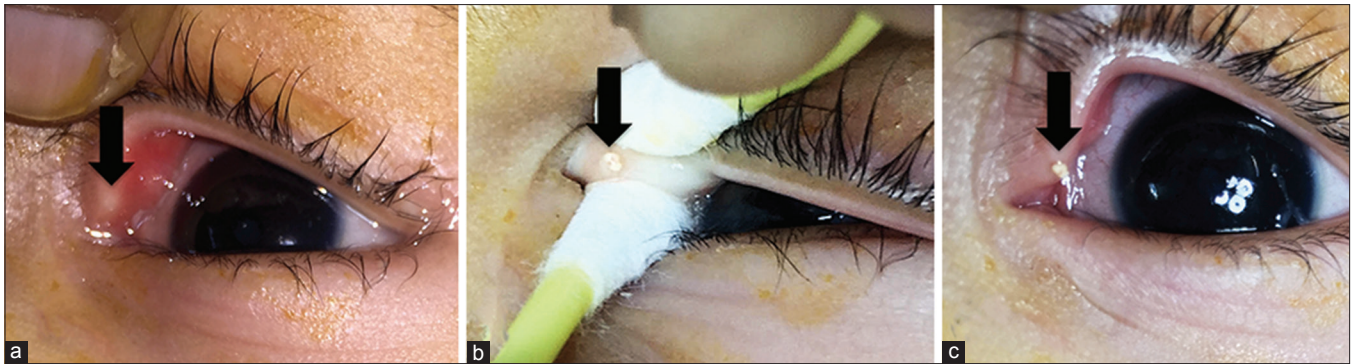


Figure 1: The upper punctum could not be clearly visualized owing to overlying discharge (black arrow in Figure 1a). On clearing the discharge, a yellowish canaliculolith (black arrow in Figure 1b) was seen plugging the pointing punctal opening. Using two cotton tip applicators, the canaliculus was squeezed, and multiple yellow, small, firm bits of granular material were expressed out (black arrow in Figure 1c)

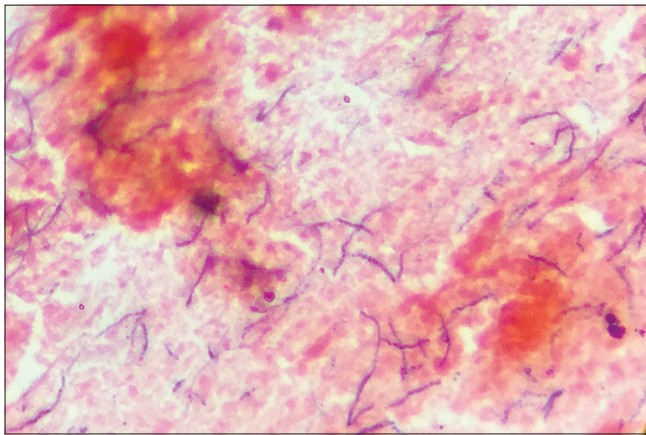


Figure 2: Photomicrograph showed irregularly arranged, delicate, branched, gram-positive filaments of *Actinomyces israelii* (Gram's stain, x100)

that canaliculitis masquerades as. Typically, epiphora, excessive discharge, and conjunctivitis along with eyelid swelling that does not respond to conventional treatment should raise the clinical suspicion of canaliculitis.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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