

Long-term Outcome of Incision and Curettage Treatment in Patients with Lacrimal Gland Ductulitis

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Purpose: To describe the effects and long-term outcomes of incision and curettage treatment in patients with lacrimal gland ductulitis.

Methods: Twenty-four patients (24 eyes) with lacrimal gland ductulitis who were treated at Saevit Eye Hospital from June 2010 to November 2016. All patients underwent incision and curettage through the lacrimal ductule, and granules or concretions were removed. After the procedure, oral and topical antibiotics, oral anti-inflammatory agent were used for a week. Clinical presentations of the patients were analyzed. The resolution of symptoms and inflammatory signs and recurrence were evaluated more than 12 months after the procedure including telephone follow-up by a specialist nurse.

Results: Common symptoms were a painful, swelling mass with mucous discharge (17 eyes) and conjunctival injection (7 eyes) at the lateral canthal area. During the procedure, 22 patients (91.7%) had typical sulfur granule of *Actinomyces*, and 10 patients (41.7%) had many cilia in the expressed debris from the ductule. Twenty-three of 24 patients had resolution of symptoms after the procedure and all but one patient (95.8%) showed no recurrence.

Conclusions: Incision and curettage is a simple and less invasive procedure that may be considered as a first treatment option for lacrimal gland ductulitis. Furthermore, incision and curettage of the affected lacrimal ductule has been shown to be effective at minimizing long-term recurrence of lacrimal ductulitis.

Key Words: Actinomyces, Curettage, Lacrimal apparatus, Lacrimal gland ductulitis

Lacrimal gland ductulitis is an inflammatory disorder characterized by infection of the lacrimal gland ductules [1,2]. Clinical manifestations of lacrimal gland ductulitis have been reported, but effective management approaches have not been well defined. *Actinomyces*—the probable

pathogen of lacrimal gland ductulitis—are anaerobic bacteria first associated with the etiology of lacrimal canaliculitis by Ellis et al. [3] in 1854. Since the chief symptoms of lacrimal gland ductulitis mimic other common ocular conditions (e.g., canthal mass, conjunctival injection and mucous discharge at the lateral canthal area), this condition is often misdiagnosed as chronic conjunctivitis, hordeolum or lacrimal gland ductal cyst (known as dacryops) infection.

Dacryops infection is a secondary infection of a pre-existing sac which primarily occurs by bacterial infection followed by enlargement of the lacrimal gland ductule. The same procedure used to treat dacryops infection and

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lacrimal gland ductulitis is surgical excision of the affected cysts and ductule of the lacrimal gland [1,2]. However, despite different pathogenesis of these disorders, the therapeutic approach for dacryops infection was applied to treat

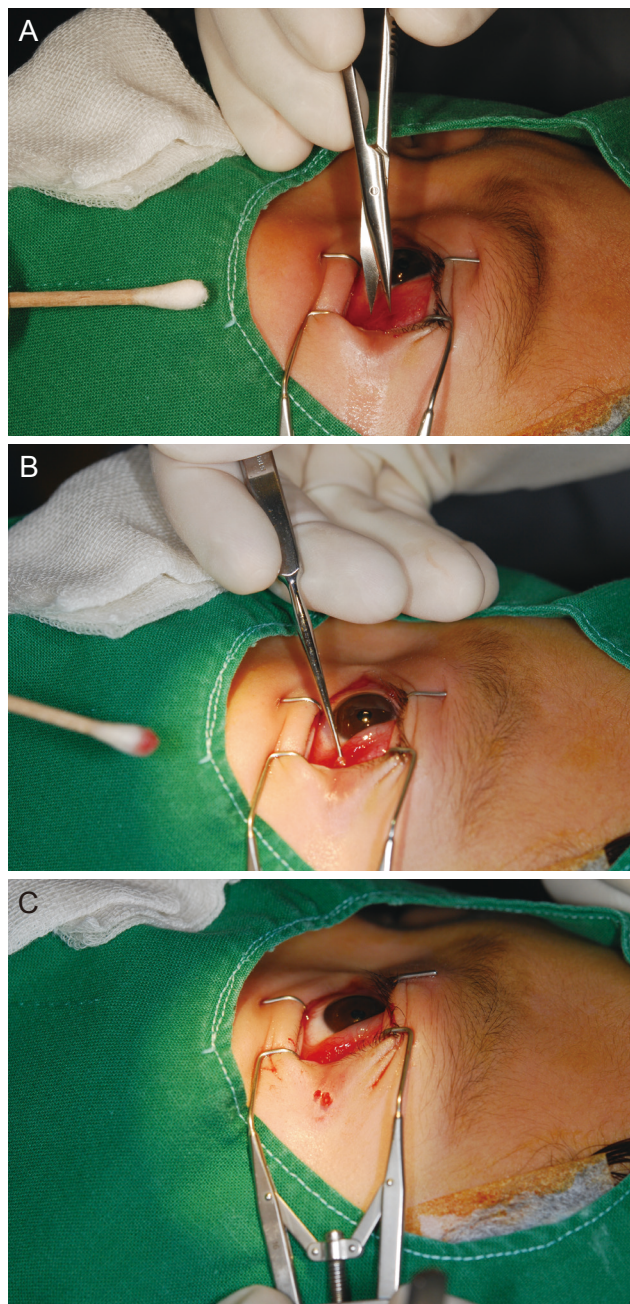


Fig. 1. Patient with lacrimal gland ductulitis underwent incision and curettage. (A) An extended wall of the lacrimal gland ductile was incision and expanded using a Westcott stitch scissors (Katena K4-4100). (B) A Meyerhoefer chalazion curette 1.75 mm (Stephens S4-1005) was inserted into the lacrimal gland ductule and removed until no more contents. (C) Sulfur granules are suspected. Informed consent was obtained from all study participants.

lacrimal gland ductulitis. Complete excision of the ductule or cysts for treatment of lacrimal gland ductulitis is a complicated and time-consuming procedure.

Under the supposition that both lacrimal gland ductulitis and lacrimal canaliculitis are caused by *Actinomyces* infection, the authors of this study attempted incision and curettage (I&C)—a simple and effective surgical intervention to manage lacrimal canaliculitis—to treat lacrimal gland ductulitis. Our study aimed to characterize the effectiveness of I&C instead of complete excision of the lacrimal gland ductule in patients diagnosed with lacrimal gland ductulitis, with a long-term follow-up of more than 12 months.

Materials and Methods

This study retrospectively reviewed patients who diagnosed with lacrimal gland ductulitis at our hospital from June 2010 to November 2016. Chart reviews were conducted to determine age, sex, clinical symptoms, onset site, ophthalmologic findings and histopathologic findings at diagnosis. Furthermore, we reviewed treatment approaches and outcomes. This study was approved by the institutional review board of Saevit Eye Hospital (SVEC 201908-003-01). Informed consent was waived due to the retrospective nature of the study.

We thoroughly examined the lacrimal puncta, lacrimal canaliculi, eyelids, lateral canthus, medial canthus, lacrimal glands, lacrimal gland ductule and conjunctiva in all patients at the time of the first visit. Patients with mucous discharge and conjunctival injection at the lateral canthus, redness and swelling of lacrimal gland ductule and discharge from the lacrimal ductule were diagnosed of having lacrimal gland ductulitis. No patients had a history of symptoms suggesting the presence of lacrimal gland ductal cysts and all underwent I&C of the lacrimal gland ductule under local anesthesia.

Subcutaneous and subconjunctival area in affected lateral canthus was anesthetized by injecting the combined solution of 2% lidocaine with 1 : 100,000 epinephrine. The eye was exposed using the eye speculum (K1-5675; Katena Products Inc., Denville, NJ, USA), and then the ductule wall of the enlarged lacrimal gland was partially excised and widened using Westcott stitch scissors (K4-4100, Katena Products Inc.). Subsequently, curettage was applied to

completely remove infectious materials from the lacrimal ductule by inserting a Meyerhoefer chalazion curette, 1.75 mm (S4-1005; Stephens Inc., Lexington, KY, USA) into the lacrimal gland ductule (Fig. 1A-1C). After surgery, antibiotic eye drops (Cravit, levofloxacin 5 mg/mL; Santen Pharmaceutical, Osaka, Japan), steroid eye drops (Flumetholon 0.1, Santen Pharmaceutical), oral antibiotics (Augmentin 625 mg, amoxicillin 500 mg and clavulanate potassium 125 mg; Il Sung Pharmacia, Seoul, Korea), and an oral anti-inflammatory agent (Varidase, streptodornase 2500 IU and streptokinase 10000 IU; SK Chemical, Seoul, Korea) were given to all patients during the first postoperative week. Remission was defined as complete resolution of all clinical symptoms and signs after the surgery until the last follow-up. Patients follow-up included outpatient visits and

telephone calls by a special nurse for a period of at least 12 months.

Results

Twenty-four patients (24 eyes), including 11 males (11 eyes) and 13 females (13 eyes), with a mean age of 37.0 ± 18.0 years (range, 8 to 83 years) were included in the study (Table 1). Patients visited our hospital with a chief complaint of a painful mass with mucous discharge (17 eyes) and conjunctival injection at the lateral canthal area (7 eyes) (Fig. 2A). All patients were initially managed with local and oral antibiotic therapy due to a suspicion of chronic conjunctivitis, hordeolum, dacryops infection and others, however, were eventually referred to our hospital

Table 1. Demographics and clinical presentation and treatment of 24 cases of lacrimal gland ductulitis

Case no.	Sex/age (yr)	Affected area	Symptom	I&C and histologic finding	Surgical outcome/ FU period (mon)
1	F/25	Left	Painful lid swelling	Sulfur granule	No recurrence/89
2	M/43	Right	Painful lid swelling	Sulfur granule	No recurrence/86
3	F/13	Left	Lid mass	Sulfur granule, cilia	No recurrence/80
4	M/42	Right	Lid mass	Sulfur granule, cilia	No recurrence/79
5	F/14	Left	Lid mass	Sulfur granule, cilia	No recurrence/79
6	M/39	Left	Painful lid swelling	Sulfur granule, cilia	No recurrence/77
7	M/44	Right	Conjunctival injection	Sulfur granule, cilia	No recurrence/76
8	M/41	Left	Painful lid swelling	Sulfur granule, cilia	No recurrence/75
9	M/8	Left	Conjunctival injection	Sulfur granule, cilia	No recurrence/72
10	F/31	Left	Conjunctival injection	Sulfur granule	No recurrence/71
11	F/31	Right	Pus discharge	Granuloma	No recurrence/68
12	M/50	Left	Conjunctival injection	Sulfur granule, cilia	No recurrence/66
13	F/20	Left	Lid mass	Sulfur granule	No recurrence/40
14	F/16	Right	Painful lid swelling	Sulfur granule, cilia	No recurrence/35
15	M/34	Right	Conjunctival injection	Sulfur granule	No recurrence/25
16	M/66	Right	Conjunctival injection	Sulfur granule	No recurrence/25
17	F/22	Left	Painful lid swelling	Sulfur granule	No recurrence/12
18	F/42	Left	Lid mass	Sulfur granule	Recurrence/15
19	M/83	Right	Lid mass	Sulfur granule	No recurrence/20
20	F/33	Right	Pus discharge	Sulfur granule	No recurrence/17
21	F/64	Left	Conjunctival injection	Granuloma	No recurrence/19
22	F/51	Left	Pus discharge	Sulfur granule, cilia	No recurrence/16
23	F/32	Right	Pus discharge	Sulfur granule	No recurrence/15
24	M/51	Right	Mucous discharge	Sulfur granule	No recurrence/15

I&C = incision and curettage; FU = follow-up.

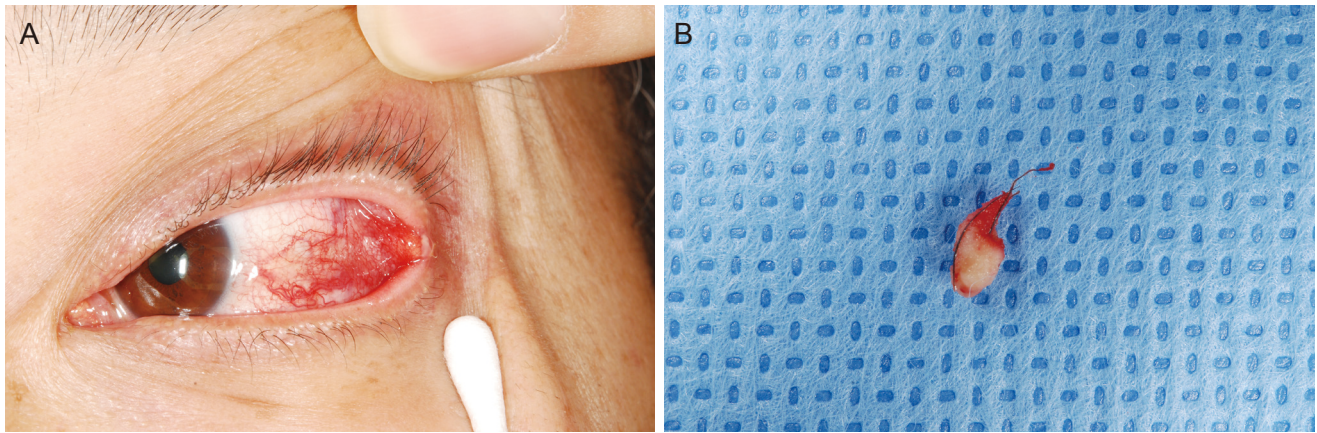


Fig. 2. Patient with lacrimal gland ductulitis and dacryolith. (A) Patient with lacrimal gland ductulitis referred with painful lid swelling, persistent temporal conjunctival injection, and chronic mucopurulent discharge from the lacrimal gland ductule. (B) Many cilia in the expressed debris from the affected ductule. Informed consent was obtained from the study participant.

because they were refractory to antibiotic therapy. All patients underwent surgical intervention by I&C of the lacrimal gland ductule under local anesthesia. Large granulation tissue and dacryolith visible to the naked eye were observed intraoperatively in 22 (91.7%) of 24 eyes and many cilia around dacryoliths in 10 (41.7%) of 24 eyes (Fig. 2B). Based on histopathologic examination, 22 eyes (91.7%) had dacryoliths formed by sulfur granules caused by *Actinomyces* infection, and 10 eyes (41.7%) had necrotic tissue consisting of ciliary follicles. After the procedure, 23 (95.8%) of 24 eyes showed resolution of the symptoms and signs of lacrimal gland ductulitis at the last follow-up. There were no any postoperative complications. The average length of follow-up was 48.3 ± 29.2 months (range, 12 to 89 months). All patients were followed up via outpatient visits and telephone calls and all but one patient (95.8%) showed no recurrence. The relapsing patient experienced the relapse on the first week after I&C, and then underwent excision of affected lacrimal gland ductule and biopsy revealing the formation of granulation tissue and dacryolith. Thereafter complete remission was achieved without relapse.

Discussion

Lacrimal gland ductulitis is an inflammatory disorder caused by infection of the lacrimal gland ductules; common clinical manifestations are swelling of the lacrimal gland ductule, conjunctival injection and a painful mass

associated with mucous discharge from the lateral canthus. This condition is characterized, in part by the formation of a yellowish stone or granulation tissue in the lacrimal gland ductule [1,2]. The subjects of this study had typical clinical signs of lacrimal gland ductulitis. However, some patients had mild symptoms and only conjunctival injection and mucous or purulent discharge from the lacrimal ductules without clear swelling or redness of the lacrimal gland ductule in slit-lamp biomicroscopy.

The etiology of lacrimal gland ductulitis still remains unclear but is presumed to be caused by *Actinomyces* infection of the lacrimal gland ductules [1,2]. The *Actinomyces* species—gram-positive, anaerobic bacteria that colonize the human mouth—may develop a chronic granulomatous infection through direct spread to the normal flora of the mouth. The *Actinomyces* species have been identified as common pathogens causing lacrimal canaliculitis, conjunctivitis, dacryocystitis, keratitis and endophthalmitis [4-8]. Lacrimal gland ductulitis can be very difficult to differentiate from dacryops infection. Dacryops is formed and enlarged by excessive production of IgA due to inflammation or trauma around the lacrimal ductules, contraction or destruction of the nerve root of the lacrimal ductules, and weakening of the ductal walls [9-12]. A lacrimal ductal cyst presents as asymptomatic swelling in the lateral portion of the upper eyelid and appears as a bluish white, soft, mobile, translucent mass [13-15].

Dacryops infection is a very rare disease and there are only 5 cases reported in literatures listed on PubMed. Because dacryops is a cystic form, there is no discharge. Da-

cryops infection may be associated with sudden pain and/or a gradual increase in mass size, and can be managed with complete excision of the cyst or marsupialization, but complete excision of the cyst is the preferred approach of treatment with no recurrence [12-17]. Differential diagnosis between dacryops infection and lacrimal gland ductulitis is easily lead by the history of lacrimal ductal cyst, but challenging in those without this history. Also histological difference is able to distinguish, but only after surgery (e.g., local or complete excision of the cyst). Histopathological difference of dacryops infection and lacrimal gland ductulitis is often difficult since, in chronic cases, the cyst wall consists of pseudostratified nonkeratinized, basal columnar, and superficial myoepithelial cell layers, double-layered lining of non-ciliated columnar cells and a single layer of cells [9-13,16,18,19]. Lee et al. [1] reported a case of pseudoepitheliomatous hyperplasia in the invaded ductule as one of the histologic features of lacrimal gland ductulitis. However, this new feature seems insufficient to distinguish between the two disorders. Because of difficulties in differential diagnosis, lacrimal gland ductulitis may have often be misdiagnosed as dacryops infection.

Lee et al. [1] and Hay-Smith and Rose [2] documented 7 and 12 patients with lacrimal gland ductulitis, respectively, and successfully treated them by performing complete excision of the affected lacrimal gland ductule. In addition, anatomical pathological examination confirmed the presence of sulfur granules and *Actinomyces*. The pathophysiology and histologic features of lacrimal gland ductulitis resemble those of lacrimal canaliculitis—an infection and inflammation of the lacrimal canaliculi. Lacrimal canaliculitis is treated with I&C of the canaliculus, a simple procedure that is less invasive compared with complete excision [20,21]. Based on these observations, the authors of this study performed I&C of the lacrimal gland ductules to manage lacrimal gland ductulitis of 24 eyes. During I&C, we detected sulfur granules visible to the naked eye in 22 of 24 eyes and a number of cilia in the expressed debris from the ductule in 10 eyes. Since infection by *Actinomyces* usually occurs because of direct extension, cilia may be the initial nidus for the development of lacrimal gland ductulitis. Although *Actinomyces* is suspected to be the causal organism for both lacrimal canaliculitis and lacrimal gland ductulitis, cilia are frequently found in lacrimal gland ductulitis, unlike the rare presence of eyelashes in lacrimal canaliculitis. This might be because cilia at the

conjunctival fornix shift toward the horizontal lacrimal ductule in the lateral canthus by blinking because the punctum opens to a vertical orientation while the lacrimal ductule runs horizontally.

In this regard, Lee et al. [16] suggested that cilia may act as a nidus for lacrimal ductular inflammation or cyst formation as eyelashes shift toward the horizontal lacrimal ductule in the lateral canthus. Due to the retrospective nature of the analysis, this study was limited by the relatively small sample size and lack of a control group for comparison. Our study was meaningful in that it successfully achieved complete remission in 23 of 24 eyes by performing less invasive I&C compared with complete excision of the lacrimal gland ductule, and had no recurrence during follow-up (range, 12 to 89 months).

We concluded that, lacrimal gland ductulitis should be considered when swelling of the lacrimal gland ductule, chronic conjunctival injection and a painful mass associated with mucous discharge from the lateral canthus are present. I&C is a simple and less invasive procedure that may be considered as a first treatment option for lacrimal gland ductulitis. Furthermore, I&C of the affected lacrimal ductule has been shown to be effective at minimizing long-term recurrence of lacrimal ductulitis.

Conflict of Interest

No potential conflict of interest relevant to this article was reported.

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