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# Outcomes of lacrimal probing surgery as the first option in the treatment of congenital dacryocystocele

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***Abstract***

**Purpose:** To determine the demographic and clinical characteristics of newborn patients who underwent lacrimal probing surgical intervention with or without the marsupialization of intranasal cysts as the primary management for dacryocystocele treatment.

**Methods:** Data from the medical charts of 350 infants who underwent lacrimal probing surgery due to nasolacrimal duct obstruction were reviewed retrospectively. Ten newborn patients with a naive diagnosis of congenital dacryocystocele were included in the study. Congenital dacryocystocele diagnosis was based on a triad of swelling in the inner canthal region, a bluish appearance, and epiphora. Lacrimal probing surgery accompanied by nasal endoscopy was planned for all patients as the first treatment option.

**Results:** The mean age of the patients was 24.90 ± 7.15 days, with a range of 6–85 days. A total of 10 patients were included, comprising seven females and three males. The mean postoperative follow-up period was 38.7 ± 24.41 months. Five patients had left, four patients had right, and one patient had bilateral dacryocystocele. Seven eyes of the six patients had uncomplicated dacryocystocele, while the remaining patients had dacryocystocele with complications of dacryocystitis and/or preseptal cellulitis. All patients had intranasal cysts. All patients underwent one session of lacrimal probing surgery under general anesthesia; all with successful outcomes. Four patients with additional dacryocystocele-associated complications underwent combined intranasal marsupialization of the cyst wall.

**Discussion:** Lacrimal probing surgery ± intranasal marsupialization of the cyst wall as a first treatment option can be effective for both congenital dacryocystocele and/or congenital dacryocystocele plus associated complications and provide complete resolution of dacryocystocele-related symptoms.

***Keywords:*** dacryocystocele, intranasal cyst, intranasal marsupialization, lacrimal probing

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## Introduction

Congenital dacryocystocele (CDC), an uncommon variant of congenital nasolacrimal duct obstruction (NLDO), is a drainage disorder of the lacrimal system accompanied by epiphora and bluish swelling in the inner canthal region that develops during the first 12 weeks of life or in newborns.[1–3] It has been reported that CDC develops as a result of the anatomical obstruction of the Hasner valve, where the nasolacrimal canal opens to the inferior meatus of the nose with the simultaneous functional obstruction of the Rosenmuller valve, where the common canaliculus is discharged into the lacrimal sac.[3] As a result, tears, mucus, amnion fluid, and infected materials are stuck in the lacrimal system and cause

lacrimal sac distension, which presents as a bluish hard mass under the medial canthal tendon with symptoms of NLDO at birth.

The optimal treatment strategy for CDC remains controversial. Initial conservative treatment can be a viable option in the absence of complications, such as dacryocystitis, preseptal cellulitis, and respiratory distress. Bilateral CDC is properly important because the nose is the only airway for infants to breathe.[4] Airway compromise may happen acutely with bilateral swelling. Thus, in case of respiratory distress signs, immediate surgical intervention should be performed.[4] It has been shown that conservative treatment may be sufficient because of the high rate of spontaneous resolution, similar to that of congenital NLDO.[5] However, systemic antibiotic treatment and/or lacrimal probing surgery should be applied in case of complications. However, lacrimal probing surgery can also be thought of as the first choice of treatment for CDC without complications or secondary infections.

The purpose of this study was to determine the demographic and clinical characteristics of patients with CDC and investigate the efficacy of lacrimal probing surgery ± intranasal marsupialization as the first choice of treatment for both CDC alone and/or CDC associated with complications. According to a literature review, our study might be the first to address lacrimal probing surgery ± intranasal marsupialization of the cyst wall for the primary management of CDC alone and/or CDC associated with complications.

## Methods

The data of 350 infant patients who underwent lacrimal probing surgery due to NLDO in a tertiary eye center between January 2011 and December 2018 were analyzed retrospectively. The medical records of 10 patients who underwent lacrimal probing surgery due to CDC were also reviewed and included in the study. Demographic data, mode of delivery, birth weight, maternal age, week of delivery, postnatal hospitalization (duration of the hospital stay after birth), the additional diagnosis of CDC complications, and preoperative and postoperative ophthalmologic examination findings were recorded (see Table 1).

CDC diagnosis was based on the triad of swelling in the inner canthal region, a bluish appearance, and eye discharge or epiphora (not mandatory) (see Figure 1(a)). Dacryocystitis was co-diagnosed with CDC in cases with redness, pain, and warmth on palpation in addition to the triad (see Figure 1(b)). Since the bluish appearance is masked by the inflammation in cases with secondary dacryocystitis, the CDC diagnosis was confirmed by the ultrasonography representing well-defined cystic swelling. The co-diagnosis of preseptal cellulitis was based on eyelid edema and erythema in addition to former clinical symptoms. Previously started systemic antibiotic treatments of patients with dacryocystitis or preseptal cellulitis by pediatricians were continued as recommended. Lacrimal probing surgery was planned for all patients as the primary management. The operation records were also reviewed, and pre- and perioperative difficulties and peri- and postoperative complications were assessed.

### *Surgical procedure*

All the patients underwent lacrimal probing surgery accompanied by a small scoped nasal endoscopy (EG-530 N2; Fujifilm Co., Saitama, Japan) following inferior turbinate vasoconstriction under general anesthesia by the same experienced surgeon. After we dilated the upper and lower punctum and canaliculi, the common canaliculus was passed with a 00/00 numbered lacrimal probe (0.55 or 0.60 mm Bowman probe, Lachrymal BOWMAN 02–03; Inami, Tokyo, Japan) through both the lower and upper punctum, and the lacrimal sac was reached. Finally, we passed the probe through the nasolacrimal duct, rotated 90°, advanced vertically, and passed the blocked Hasner valve to reach the inferior meatus. The lacrimal probes were removed after metal-to-metal contact was felt with another lacrimal probe inserted vertically through the nostril. We washed the lacrimal sac with saline to provide lacrimal sac drainage. Irrigation was applied with a lacrimal cannula, and after we observed that the cotton-tipped rods placed in the nasal cavity were wet, the lacrimal probing surgery was terminated.

For sufficient marsupialization, we used a 19-gauge angled MVR Lance (Feather Safety Razor, Osaka, Japan) and small through-cutting forceps, straight shaft, upward (No.10270602; Nagashima Medical Instruments, Tokyo, Japan). We then safely made an incision at an appropriate site in the inferior meatus without damaging other parts. We performed lacrimal sac massage to achieve complete drainage and thorough

**Table 1.** Demographic and clinical characteristics of the patients.

| Patient | Age (day) | Gender | Side | Type of delivery | Birth weight (Gram) | Birth week | Postnatal hospitalization (day) | Mother age (year) | Number of births | Surgical procedure | Control | Medical* therapy | Additional diagnosis | Follow-up duration (month) |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| 1 | 13 | F | L | C/S | 2950 | 36 + 2 | 5 | 35 | 4 | Probing | Normal | Yes | Dacrocystitis | 32 |
| 2 | 6 | F | L | C/S | 3050 | 37 + 6 | 0 | 30 | 2 | Probing | Normal | No | No | 60 |
| 3 | 85 | M | L | N | 3100 | 39 + 2 | 15 | 29 | 2 | Probing | Normal | Yes | Dacrocystitis and preseptal cellülitis | 72 |
| 4 | 12 | F | R + L | C/S | 3750 | 39 + 0 | 1 | 25 | 3 | Probing | Normal | Yes | No | 27 |
| 5 | 15 | F | R | C/S | 3150 | 38 + 2 | 0 | 27 | 1 | Probing | Normal | No | No | 9 |
| 6 | 24 | F | R | N | 3250 | 40 + 2 | 3 | 26 | 2 | Probing | Normal | No | No | 32 |
| 7 | 70 | M | R | N | 3200 | 39 + 5 | 5 | 28 | 3 | Probing | Normal | Yes | Dacrocystitis | 63 |
| 8 | 6 | F | R | N | 3110 | 38 + 4 | 2 | 22 | 3 | Probing | Normal | No | No | 12 |
| 9 | 10 | M | L | N | 3300 | 38 + 4 | 2 | 33 | 2 | Probing | Normal | No | No | 12 |
| 10 | 8 | F | R | C/S | 3500 | 39 + 3 | 4 | 30 | 2 | Probing | Normal | No | Dacrocystitis | 33 |

C/S, cesarean section; F, female; L, left; M, male; N, normal delivery; R, right.
*Systemic antibiotic treatment.

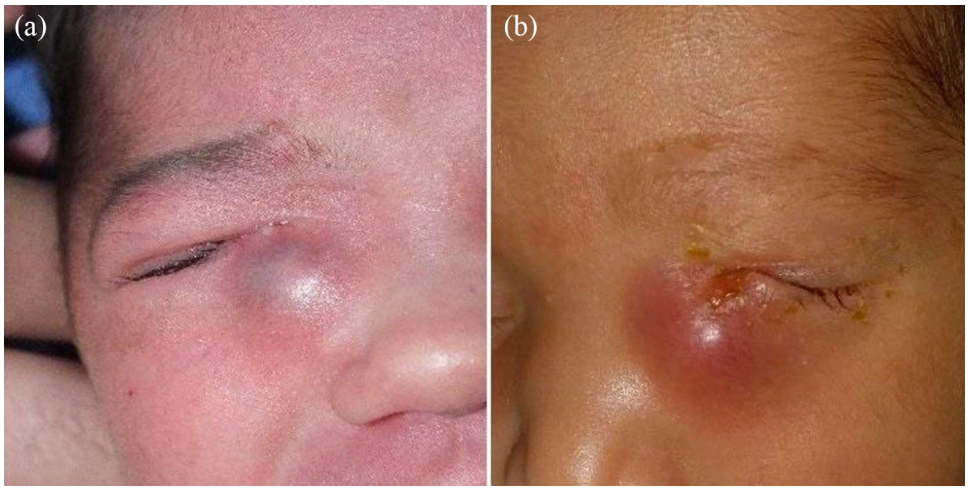


**Figure 1.** Uncomplicated congenital dacryocystocele with blueish swelling in the inner canthi (a) and congenital dacryocystocele with dacryocystitis (b).

aspiration during the procedure to prevent pus flow into the pharynx to prevent additional stress and pneumonia.

Patients with CDC alone were postoperatively prescribed a combination of dexamethasone 0.1% and netilmicin 0.3% four times a day for 1 week. Patients with complicated CDC (dacryocystitis or preseptal cellulitis) were continued with previously ordered systemic antibiotic therapy and additionally prescribed with dexamethasone 0.1% + netilmicin 0.3% as well as patients with CDC alone. The patients had regular postoperative visits on day 1, week 1, month 1, month 3, and later on yearly. The mean postoperative follow-up period was 35.20 ± 20.41 months, with a range of 9–72 months. Our success criteria were the complete improvement of swelling, watering, and discharge complaints based on ophthalmic physical examinations and information taken from the parents during regular hospital visits.

## Results

This study included 10 patients comprised of 7 females (70%) and 3 males (30%). The ages ranged from 6 to 85 days, and the mean age was 24.90 ± 7.15 days. CDC was diagnosed in 11 eyes, including 5 left eyes, 4 right eyes, and 1 bilateral involvement. Clinical and demographic properties, natal and postnatal maternal and newborn features, medical treatments, and surgical outcomes are presented in Table 1. Patients with an additional diagnosis of CDC complication had a longer postnatal hospitalization compared to patients with CDC alone (7.25 ± 3.60 days and 1.33 ± 0.56 days, respectively). We did not observe any association between additional diagnoses and age, gender, type of delivery, birth weight, maternal age, or the number of births. Six patients (7 eyes) without any previous massage or conservative treatment were directly admitted to our clinic with the diagnosis of CDC alone. These patients received a lacrimal probing surgery as the primary management. Three patients had associated dacryocystitis, and one patient had associated dacryocystitis plus preseptal cellulitis secondary to CDC. These four patients were previously prescribed systemic antibiotic treatments by their pediatricians due to misdiagnoses of CDC-associated complications. They were consulted to our clinic due to nonimproving symptoms, despite the systemic treatments. They were then co-diagnosed with CDC and admitted with their ongoing treatments and subsequently given a combined intranasal marsupialization of the cyst wall and lacrimal probing surgery as the primary management.

Intranasal cysts were detected in all cases during perioperative nasal endoscopy. However, marsupialization was performed in only complicated cases related to a larger size and increased contents. All patients had successful lacrimal probing surgeries, including four patients who had a combined intranasal marsupialization of the cyst wall due to complicated CDC with dacryocystitis and preseptal cellulitis (Figures 2(a) and (b) and 3(a) and (b)). Controlled perioperative hemorrhages were observed in only two patients. We did not

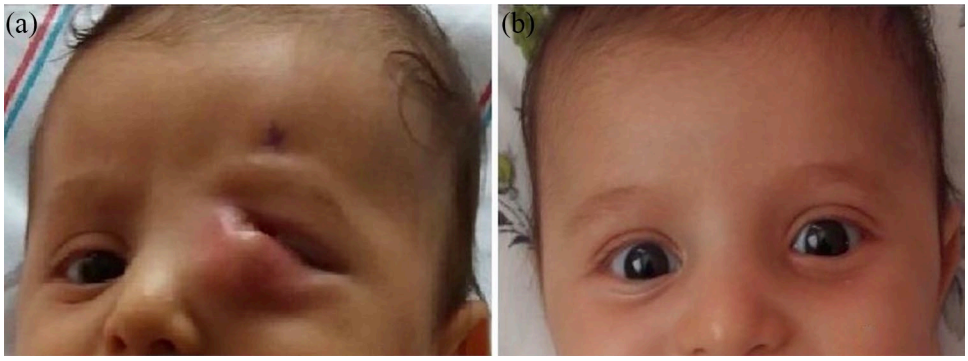


**Figure 2.** Dacryocystocele combined with dacryocystitis and preseptal cellulitis (a) and postoperative first month view of the same case (b).

note any other perioperative or postoperative complications. Following the probing surgery, complaints of swelling, watering, and discharge completely improved in all patients. Thus, a certain clinical and functional success was achieved.

## Discussion

Between 35% and 73% of newborns have imperforated nasolacrimal ducts, but most of these open spontaneously in the first postnatal week, so CDC is a rare disease.[5,6] Prenatally diagnosed dacryocystoceles had an incidence of 0.43% in the United States with the highest ratio in the early third trimester and the incidence decreases thereafter. This is followed by complete recovery in 76% of these infants at birth, and the gestational age at delivery is a significant determinant for the postnatal persistence of dacryocystocele.[7] With a CDC diagnosis, physicians should refrain from additional postnatal diagnostic methods and treat the potential postnatal complications.[8] There are no precise results concerning CDC prevalence; however, a study conducted in the United States reported a ratio of 1 per 3,884 newborns, while a study from the United Kingdom reported a ratio of 1 per 18,597.[3,9] In another study, dacryocystocele was reported as 0.1%, regardless of the requirement for probing surgery in patients with NLDO.[10] We think other possible causes for prevalence discrepancies might include poor coordination between newborn pediatric clinics and eye clinics, late or misdiagnoses, insufficient statistical medical records, and spontaneous or therapeutic postnatal recovery of the disease. Previous studies have reported that CDC is more common in female patients; findings similar to what we observed with a female dominance in our study.3,9,11–13 Dacryocystoceles generally present with unilateral subcutaneous swelling in the lacrimal sac area and are related to dilatation of the nasolacrimal duct. Moreover, in some cases, dacryocystoceles may also present with large intranasal cysts.[14] We also observed intranasal cysts in the nasal endoscopic examination, similar to reports in the literature with various incidence rates.[1,3,9] Unilateral dacryocystocele predominance was noted in our study, which concurs with many other studies.[3,9,15,16]

An important problem in newborn patients with dacryocystocele is that the risk of dacryocystitis occurs in up to 75% of cases.[3,17] In our study, dacryocystocele was accompanied by dacryocystitis at a rate of 36.36%. Stasis in the lacrimal sac, proximity to the sinuses, and a high submucosal lymph density and vascularity in the lacrimal sac have been reported to pose a risk for secondary infection.[3,17] In addition, another reason for secondary infections may be treatment recommendations, such as incorrect massage methods and/or an excessive amount of massage. For patients with both common canaliculus obstruction and NLDO, the incorrect application of massage to the stretched, filled, and bulging pouch may damage the sac wall and cause the sac contents to leak into the subcutaneous area.

Theoretically, there are various treatment options for the CDC, including observation, digital massage combined with antibiotic therapy, intranasal marsupialization of an infected cyst, and lacrimal probing surgery. However, there is still confusion in the literature regarding the time and duration of these treatment options. Many authors recommend massage and antibiotic therapy for uncomplicated dacryocystocele.2,11,13,16–20 Although it was not statistically significant, Davies and

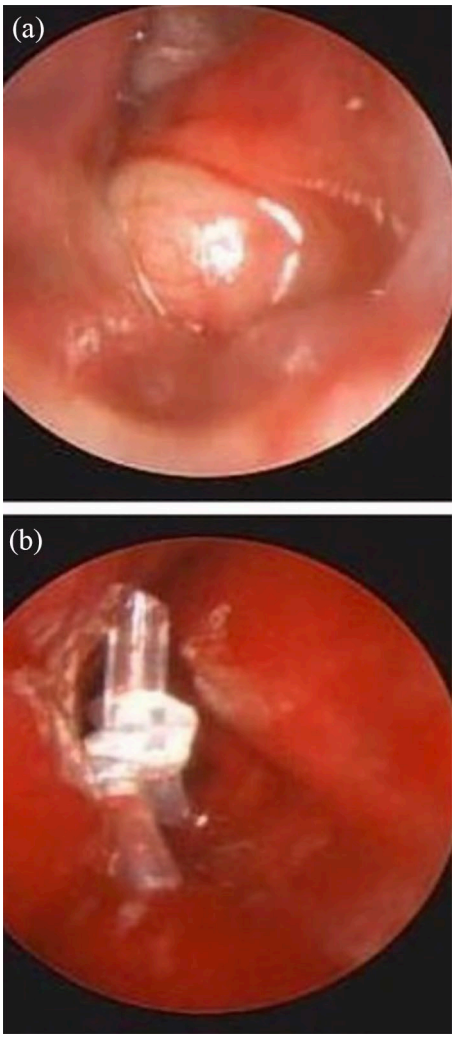


**Figure 3.** Intraoperative endoscopic images of left dacryocystocele before the marsupialization (a) and after the marsupialization (b).

colleagues[9] reported that the incidence of surgical intervention was higher in cases with dacryocystocele plus dacryocystitis than in cases with dacryocystocele alone. Some studies have reported that dacryocystocele was resolved with conservative management alone, without surgical interventions.[3,5] CDC may be misdiagnosed as secondary infections of dacryocystitis and/or preseptal cellulitis, especially by pediatric clinics, which may lead to late diagnosis and unnecessary mistreatments. Therefore, it is essential to build strong coordination between newborn pediatric clinics and ophthalmology clinics for timely and correct treatments. Misdiagnoses and late hospital admissions may cause complications and delay proper treatments, especially surgical probing intervention. We think that misdiagnoses and late admissions resulted in the relatively older mean age of newborn patients in our study compared to previous studies.[3,9]

Different results are reported in the literature regarding treatment approaches. Lee and colleagues[21] stated that most (74.1%) uninfected CDC can be resolved by conservative management; lacrimal probing was needed in only five (18.5%) persistent cases. Schnall and Christian[18] reported a 76% improvement and opening of the obstruction after 6 days of medical treatment in non-infected dacryocystocele; the remaining 24% were reported as persistent obstructions, and probing surgery was required despite 2 weeks of treatment. Wong and Vander Veen[12] stated that only 22% of patients treated with medical therapy improved; 78% of them required surgical treatment. Paysee and colleagues[20] stated that both methods (conservative treatment and lacrimal probing surgery) were successful, but they treated 97% of the patients with surgical methods. Recent studies suggest endoscopic evaluation in the diagnosis of CDC, concluding that nasal endoscopy surgery alone and/or antibiotic therapy had relatively safe and successful consequences for the treatment of CDC.[22–24] Unlike our results, in a series of 23 cases with CDCs, only 43 % (3/7) of the uncomplicated CDC patients recovered with conservative therapy.[25] Similar to our results, Levin and colleagues[26] documented that approximately 1/4 of the 19 CDC cases with secondary dacryocystitis improved with partial resolution of the infection following antibiotic treatment, but the patients needed lacrimal probing + marsupialization for absolute recovery. Suggesting our findings, another series of CDC with secondary infection reported that only 20% (2/10) of the cases recovered without surgical management, but the remaining 80% of the cases had a lacrimal probing for a complete improvement.[27]

The previous major series of CDC marsupialization reporting the treatment details in CDC cases were compared in Table 2, including this study. Consistent with our results, all studies in this table reported 100% success of probing with marsupialization in CDC cases regardless of secondary infection. This study and the studies conducted by Ali and colleagues[14] and Levin and colleagues[26] had relatively small sample sizes compared with the remaining studies because these studies included almost only the cases with intranasal cysts. Therefore, conservative management (only 1 patient in Levin and colleagues' study

**Table 2.** Treatment properties of the congenital dacryocystocele regarding previous reports and the present study.

| Study | Number of the patients (eyes) | Conservative Tx/failure (%) | Acute dacryocystitis (%) | Probing* (failure, %) | Intranasal cysts (U/B) | Probing + marsupialization** (success, %) |
|---|---|---|---|---|---|---|
| Mansour and colleagues | 54 | 0***/- | 40 (74) | 45 (0) | 6/NA | 3 (100) |
| Paysse and colleagues | 22 (30) | 0/- | 12 (55) | 30 (1) | 9/7 | 1 (100) |
| Levin and colleagues | 25 | 1/0 (0) | 19 (85) | 0 (-) | 20/4 | 24 (100) |
| Becker and colleagues | 27 (29) | 29/26 (90) | 21 (78) | 26 (4) | NA | 4 (100) |
| Wong and colleagues | 42 (46) | 10/0 (0) | 28 (67) | 36 (8) | 6/2 | 8 (100) |
| Dagi and colleagues | 64 | 33/20 (61) | 28 (44) | 39 (0) | 8/7 | 12 (100) |
| Lueder and colleagues | 33 | 17/0 (0) | 16 (49) | 0 (-) | 21/12 | 16 (100) |
| Ali and colleagues | 15 | NA | 4 (27) | 0 (-) | 15/0 | 15 (100) |
| Davies and colleagues | 35 (38) | 35/7 (20) | 17 (49) | 6 (0) | 4/0 | 4 (100) |
| This study | 10 (11) | 0 | 4 (36) | 0 (-) | 9/1 | 11 (100) |

B, bilateral; NA, not available; Tx, treatment; U, unilateral.
*Probing monotherapy under local and/or general anesthesia.
**Combined treatment of lacrimal probing and marsupialization.
***Six cases with spontaneous resolution.

with failure) and probing monotherapy were not performed. Fortunately, combined probing and marsupialization treatment yielded complete resolutions in all patients of these three studies. Regarding other studies, probing monotherapy demonstrated high success rates with low failures, but conservative therapy showed higher failure rates partially related to secondary infection.

Our approach was to apply probing as the primary management for all 10 patients diagnosed with dacryocystocele regardless of associated complications. However, patients with dacryocystocele and dacryocystitis have a markedly thickened cyst wall due to inflammation, and the probe opening often closes immediately after the probe is withdrawn, so intranasal marsupialization of the cyst wall is often required. Due to this difficulty, we performed intranasal marsupialization of the cyst wall combined with lacrimal probing surgery. Nearly 36% of the patients (four patients and four sides) had complicated dacryocystocele. Their medical treatments had been previously started by other clinics, but these patients were consulted to our ophthalmology clinic due to lack of improvement. The remaining six patients were noncomplicated, evaluated in our clinic, and diagnosed with CDC. All 10 patients had lacrimal probing surgery (four cases with CDC plus CDC-associated complications underwent additional intranasal marsupialization of the cyst wall), and 100% success was achieved in all these patients.

In terms of postoperative complications and recurrence, Mansour and colleagues[17] reported a 22% recurrence during a 6-month follow-up period, while Harris and DiClementi[2] determined a 25% recurrence rate. Since the lacrimal sac structure was expanded in these patients, the entrance of the nasolacrimal canal could be displaced. Therefore, blindly performed probing

may have difficulty in finding a preoperative nasolacrimal canal. In two cases, insignificant perioperative sac hemorrhages developed, and after the hemorrhages were removed by lavage, the surgery was continued. No postoperative complications or recurrences were observed in any of our cases. Nonetheless, this study has some limitations, including a small sample size, retrospective nature, and uni-centered design. Considering these limitations, new prospective studies with larger sample sizes and multicentered design should be conducted in future investigations.

In conclusion, lacrimal probing surgery with or without intranasal marsupialization of the cyst wall is an effective and successful treatment method as primary management for CDC alone or CDC plus associated complications. If the patient's general condition allows, it can prevent complications, decrease hospitalization duration, and prevent unnecessary antibiotic therapy. In addition, nasal endoscopic examination is important in patients with CDC for the detection of accompanying pathologies, including intranasal cysts. We suggest time-saving lacrimal probing surgery ± intranasal marsupialization of the cyst wall as the primary management to prevent CDC-associated complications, including dacryocystitis, preseptal cellulitis, and orbital cellulitis.

## Brief summary statement

Congenital dacryocystocele may be misdiagnosed with dacryocystitis in newborns and this might cause inappropriate antibiotic treatment. Preseptal cellulitis, orbital cellulitis, or orbital abscess can be observed in case of mistreatment. Lacrimal probing surgery can provide us a successful achievement as the first treatment of congenital dacryocystocele.

## Conflict of interest statement

The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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## Ethics statement

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Declaration of Helsinki and its later amendments or comparable ethical standards. The study was approvad by the local ethics committee (Ethics Committee of Clinical Researchers; approval date/Approval: 25.06.2019, approval code 2453). A written informed consent form was obtained from the parents or guardians of the patients to publish the patient data, including images and any additional related information.

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