

## Reappraisal of the Treatment of Congenital Dacryoceles

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### Keywords:

Congenital, Dacryoceles, Dacryocystocele, Epiphora, Lacrimal

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Received: 07-01-2021  
Accepted: 03-03-2021  
doi: 10.15713/ins.clever.64



### Abstract

**Background:** Congenital dacryoceles are a rare variant of congenital nasolacrimal duct obstruction with significant associated morbidity. Diagnosis and treatment for dacryoceles could be managed by an ophthalmologist or otolaryngologist.

**Aims:** This study aims to evaluate the efficacy of nasal endoscopy for the diagnosis and treatment of congenital dacryoceles.

**Methods:** A retrospective review of medical records for infants who presented with congenital dacryoceles between 2003 and 2016 at a university-affiliated tertiary pediatric medical center.

**Results:** The cohort included 28 patients. None had respiratory distress, and all received conservative treatment. Nasal endoscopy was performed at presentation in 18 patients. Fourteen (78%) were found to have an intranasal cyst and were treated with endoscopic marsupialization using a microdebrider. The remaining 10 patients underwent probing and irrigation under general anesthesia. Mean age at surgery was 30 days (range, 1 day–4 months). Dacryocel recurrence occurred in three patients, none of whom had undergone otolaryngologic evaluation. All three required further surgical intervention.

**Conclusion:** Otolaryngologic assessment should be considered for all neonates presenting with dacryocel due to the potential presence of intranasal cysts. If identified, intranasal cysts should be treated to prevent recurrence.

**Significance:** Otolaryngologic assessment could prevent cases of refractory dacryocel, thus reducing morbidity.

### Introduction

Congenital dacryocel is a rare (0.1%) variant of congenital nasolacrimal duct obstruction. It is characterized clinically by a bluish cystic mass below the medial canthus that presents shortly after birth.<sup>[1,2]</sup> The differential diagnosis includes dermoid cysts, hemangioma, nasal glioma, and lymphangioma. Dacryocel may also be identified sonographically during the second and third trimesters.<sup>[3,4]</sup>

The lacrimal system is first observed in embryos during the 5<sup>th</sup> week of gestation, and the lumen forms before the 10<sup>th</sup> week of gestation. Canalization of the cord is completed by 9 months. In some cases, however, complete canalization fails to occur, leading to the formation of a membranous barrier, usually at the valve of Hasner. Concomitant obstruction in the nasolacrimal system of the Rosenmuller valve proximally and the Hasner valve distally results

in dacryocel.<sup>[5]</sup> The distal obstruction can be both anatomical and functional. Distension of the lacrimal sac and compression of the lacrimal canal may cause a trapdoor-type blockage<sup>[1,3]</sup> resulting in the accumulation of fluid in the drainage system and posing a risk of secondary infection and inflammation. The dacryocel may extend intranasally, forming a cyst in the inferior meatus. As neonates are nasal breathers, nasal obstruction by an intranasal cyst may be associated with respiratory distress during feeding and sleeping. Bilateral dacryocel through any of these mechanisms can be life threatening and requires prompt diagnosis and treatment. First-line treatment of dacryocel includes topical or systemic antibiotics and digital massage.<sup>[6]</sup> Definitive treatment requires opening the nasolacrimal duct obstruction and marsupialization or excision of intranasal cysts, if present.<sup>[7,8]</sup> Simple probing and irrigation in the presence of intranasal cysts may not resolve the condition and could lead to recurrence.<sup>[9,10]</sup>

Infants with dacryocoele may present to the pediatrician, otolaryngologist, or ophthalmologist, depending on where the clinical signs occur. Therefore, awareness of this condition and its potential complications is important. The aim of the present study was to describe the 12-year experience of a university-affiliated tertiary pediatric medical center with dacryocoele and to assess the diagnostic and therapeutic value of early nasal endoscopy.

## Methods

### Patients and setting

A retrospective, non-comparative study design was used. The electronic database of a tertiary medical center was reviewed for all infants diagnosed with congenital dacryocoele between 2003 and 2016. The following data were extracted from the individual medical records: Patient demographics, findings on baseline clinical examination, treatment protocol, and outcome. The study protocol was approved by the local Institutional Ethics Committee and was conducted in accordance with the Declaration of Helsinki.

### Treatment protocol

According to our departmental protocol, all neonates who present with dacryocoele undergo a thorough clinical examination. In the absence of an apparent risk of airway obstruction, sepsis, and significant local infection, conservative management is initiated, consisting of massage and a topical antibiotic regimen (tobramycin 0.3%, 3 times daily), with close follow-up for any deterioration. If the dacryocoele does not resolve, a surgical approach is considered.

In neonates with a suspected local infection, systemic antibiotics are administered for 5 days, and the child is scheduled for intervention thereafter. Until 2014, patients were referred for preoperative otolaryngologic assessment at the discretion of the treating physician. However, a 2013 departmental audit showing an increased recurrence risk when an otolaryngologic examination was not performed prompted a change in our departmental protocol to include otolaryngologic assessment in every patient before surgical intervention.

Postoperatively, patients are followed for a minimum of 3 months. If symptoms resolve, they are discharged for continued care under community-based pediatricians. In the event of recurrence, the patient is referred back to us for further intervention.

### Surgical techniques

Probing and irrigation are performed under general anesthesia following dilation of the proximal nasal ducts with a metal probe and irrigation of the nasal ducts with a balanced salt solution. Nasal endoscopy is performed under general anesthesia following decongestion of the nasal cavities using a 2.7 mm rigid nasal endoscope. The inferior meatus is inspected, and

if a cyst is identified, its medial wall is removed with a 2.9 mm microdebrider (Medtronic, Minneapolis, MN, USA). If visualization is impossible, the inferior turbinate is medialized. Following marsupialization of the cyst, standard probing and irrigation are performed. Endoscopic visualization of saline with fluorescein under the inferior meatus through the Hasner's valve confirms a patent pathway. Data were analyzed by descriptive statistics. Data were generated with SPSS, version 21 (IBM Inc., Armonk, New York, USA).

## Results

The study cohort included 28 patients with dacryocoele, 11 (39%) male and 17 (61%) female, mean patient age at surgery was 30 days, with a range of 1 day–4 months. Five (18%) had bilateral disease. The patients were referred by their pediatrician following failure of resolution of infection after treatment with local antibiotics. At presentation, clinical examination revealed symptomatic dacryocystitis with significant local redness. None of the patients presented with respiratory compromise or systemic infection (sepsis). Treatment consisted of a course of local and systemic antibiotics followed by probing and irrigation under general anesthesia at some point during follow-up. Pre-operative otolaryngologic assessment evaluation was performed in 18/28 (64%) patients, of whom 14 (78%) were diagnosed with an intranasal cyst. Mean patient age at surgery was 30 days (range, 1 day–4 months).

The dacryocoele recurred within 2 months of surgical treatment in three patients. None of them had undergone otolaryngologic assessment at presentation. All had symptoms of local dacryocystitis without respiratory distress or sepsis. Otolaryngologic evaluation in one patient revealed an intranasal cyst which was subsequently marsupialized. Silicone tube insertion was performed in the other two patients. In one, the symptoms resolved; in the other, symptoms returned 3 years later warranting dacryocystorhinostomy. Neither patient underwent otolaryngologic evaluation at dacryocoele recurrence.

There were no events of recurrence among the 18 patients who were evaluated otolaryngologically before surgery and treated accordingly.

## Discussion

We describe a cohort of neonates who presented to a tertiary pediatric medical center with dacryocoele over a 12-year period. The female predominance (61%) is in agreement with the previous reports attributing dacryocoele to the narrower nasolacrimal ducts in females.<sup>[11]</sup> The dacryocoele was unilateral in most cases (81%), as reported by others as well.<sup>[1]</sup> Conservative treatment was ineffective in all patients, and all required surgical intervention for dacryocystitis. Pre-operative otolaryngologic assessment was performed at the discretion of the physician up to 2014, and thereafter, in all patients, as mandated by the updated departmental protocol. This practice was justified by

our finding that none of the 3/28 patients with a recurrence had undergone otolaryngologic assessment.

Intranasal cysts were observed on nasal endoscopy in 78% of the patients who underwent this procedure. All cysts were excised in the same session using microdebrider-assisted marsupialization under endoscopic guidance. None recurred. Two earlier small studies of congenital intranasal lacrimal duct cysts reported good results with this technique.<sup>[12,13]</sup>

There is currently no consensus on the best procedure for resolving dacryocystocele. Some pediatric ophthalmologists advocate placement of a probe into the nasolacrimal sac to decompress the dacryoceles externally, followed by probing and rupture of any internal component, without involving an otolaryngologist. Others suggest massage at home over several days until the dacryoceles pop, thereby releasing the blockage. If the blockage is not released, the sac is kept decompressed and vents the mucopurulent secretion externally. This delays probing and creates a cleaner system.

There are anecdotal reports by oculoplastic surgeons who successfully drained the dacryoceles ab externo using a 25 gauge needle followed by oral and topical antibiotics.<sup>[10]</sup> The parents were instructed to massage the area frequently and vigorously, and to decompress if they noticed reaccumulation. However, this procedure risks the development of necrosis of the overlying skin and formation of a fistula. Other pediatric ophthalmologists used systemic oral antibiotics and then intraoffice probing.<sup>[14]</sup>

In dacryoceles, the dual obstructions – at the valve of Hasner and the valve of Rosenmüller at the common canaliculus – trap material in the lacrimal sac that cannot be expressed with massage. We believe that probing does not create a large enough hole to express the material compared to marsupialization. In cases of infection, prompt removal may be the more important factor.

Our study is limited by the retrospective design and community-based follow-up of the patients. It is possible that some patients with late recurrences were referred to other institutions. Nevertheless, our findings highlight the significance of early otolaryngologic assessment with nasal endoscopy in the diagnosis and management of neonatal dacryoceles.

## Conclusion

Otolaryngologic assessment with nasal endoscopy is advocated in neonates presenting with dacryocystitis unresponsive to conservative treatment. If a nasal cyst is identified, microdebrider-assisted endoscopic marsupialization under general anesthesia is advocated to prevent recurrence.

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**How to cite this article:** Gabbay I, Gilony D, Pesoa Y, Avisar I, Soudry E, Friling R. Reappraisal of the treatment of congenital dacryoceles. *Clin Exp Vis Eye Res* 2021;4(1):1-32.

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