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Giant Congenital Dacryocystocele Presenting with Globe Displacement: A Rare Cause of Orbital Mass Effect in an Infant

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Abstract

Introduction: Congenital dacryocystocele is an uncommon form of lacrimal drainage obstruction in neonates. To the best of our knowledge, globe displacement secondary to a giant congenital dacryocystocele has not been previously reported. We describe a case of a giant congenital dacryocystocele complicated by acute dacryocystitis that produced significant orbital mass effect with globe displacement.

Case report: A 19-day-old female neonate presented with a bluish swelling over the right medial canthal region that had been present since birth. The lesion progressively enlarged and became complicated by acute dacryocystitis. Intravenous antibiotic therapy improved the inflammation; however, the mass remained enlarged and displaced the globe superotemporally. Computed tomography demonstrated a $17.7 \times 14.6 \times 16.0$ mm cystic lesion at the right medial canthus that was continuous with the nasolacrimal duct and exerted significant mass effect on the globe. Following resolution of the acute infection, nasolacrimal duct probing was performed, resulting in immediate decompression of the dacryocystocele and restoration of the globe to its normal position.

Conclusions: Giant congenital dacryocystoceles can present as a rare cause of orbital mass effect, resulting in globe displacement. Early recognition and timely intervention are essential to prevent serious complications associated with infection and airway obstruction.

Keywords: congenital dacryocystocele, dacryocystitis, globe displacement, nasolacrimal duct obstruction, nasolacrimal duct probing

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Introduction

Congenital dacryocystocele is an uncommon form of lacrimal drainage obstruction that typically presents in neonates as a bluish, cystic swelling inferior to the medial canthus. Although many cases respond to conservative management, those complicated by acute dacryocystitis often require systemic antibiotic therapy and surgical intervention. Ocular complications are rare. While compression-induced astigmatism has been described, to the best of our knowledge, globe displacement has not been previously reported. We describe a neonate with a giant congenital dacryocystocele complicated by acute dacryocystitis that produced marked globe displacement, highlighting an unusual manifestation of this condition and emphasizing the importance of prompt recognition and management.

Case Presentation

A 19-day-old full-term female neonate presented with a bluish, cystic swelling inferior to the right medial canthus that had been present since birth (Figure 1). She was otherwise healthy, with an unremarkable medical history. The pregnancy and perinatal course were uncomplicated, with no maternal history of gestational diabetes, intrauterine infection, or other pregnancy-related complications. The infant had no systemic symptoms, including fever, irritability, poor feeding, or respiratory distress.



Figure 1 Initial presentation at 5 days of age showing a bluish, cystic swelling inferior to the right medial canthus.

The patient initially presented to a local hospital, where conservative management with the Crigler massage was recommended. Despite this, the lesion progressively enlarged and became erythematous, with the development of yellowish discharge. She was admitted and treated with intravenous ceftriaxone (50 mg/kg/day) for 1 week, together with topical 0.5% moxifloxacin ophthalmic solution (Vigamox; Alcon Laboratories, Inc., Fort Worth, TX, USA) four times daily and chloramphenicol ophthalmic ointment at bedtime in the right eye (Figure 2A). During hospitalization, the inflammation subsided and the lesion decreased in size (Figure 2B). She was subsequently discharged and referred to Songklanagarind Hospital for further management.

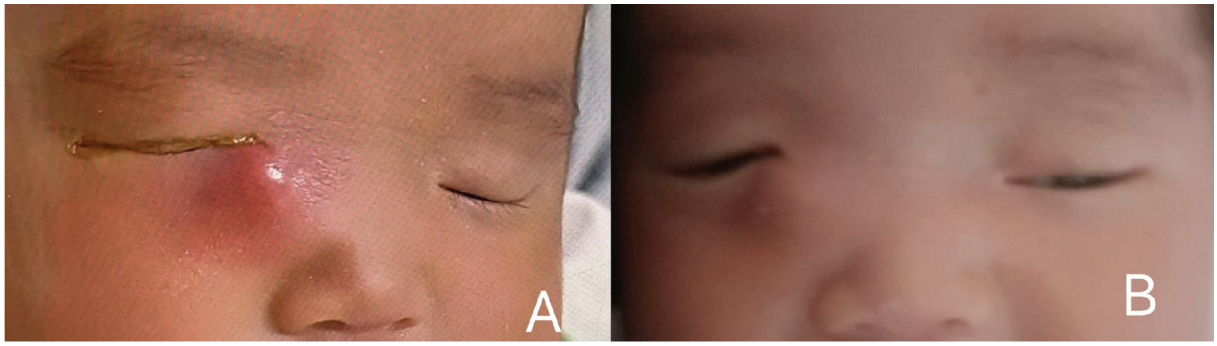


Figure 2 Clinical appearance during treatment at the local hospital. (A) At 7 days of age, on the first day of hospitalization. (B) At 15 days of age, after completion of intravenous antibiotic therapy and before referral to our institution.

At presentation to Songklanagarind Hospital at 19 days of age, the lesion had enlarged following a 5-day interruption of antibiotic therapy and caused superotemporal displacement of the right globe (Figure 3A, B). There was no evidence of respiratory distress. Intravenous ceftriaxone was restarted, and topical antibiotic therapy was continued. Given the clinical suspicion of a giant dacryocystocele complicated by acute dacryocystitis with globe displacement, orbital computed tomography (CT) was performed. Imaging demonstrated a 17.7 × 14.6 × 16.0 mm hypodense cystic lesion at the right medial canthus extending into the nasolacrimal duct, with subtle peripheral rim enhancement. The lesion exerted significant mass effect, causing compression and superotemporal displacement of the right globe (Figure 4). These findings confirmed the diagnosis of a congenital right dacryocystocele complicated by acute dacryocystitis with secondary globe displacement. Intravenous antibiotic therapy was subsequently changed to cefazolin (25 mg/kg/dose every 8 hours) to provide appropriate coverage for the most common causative organisms of acute dacryocystitis, including methicillin-susceptible Staphylococcus aureus (MSSA) and Streptococcus species, while offering a more favorable safety profile than ceftriaxone in neonates.

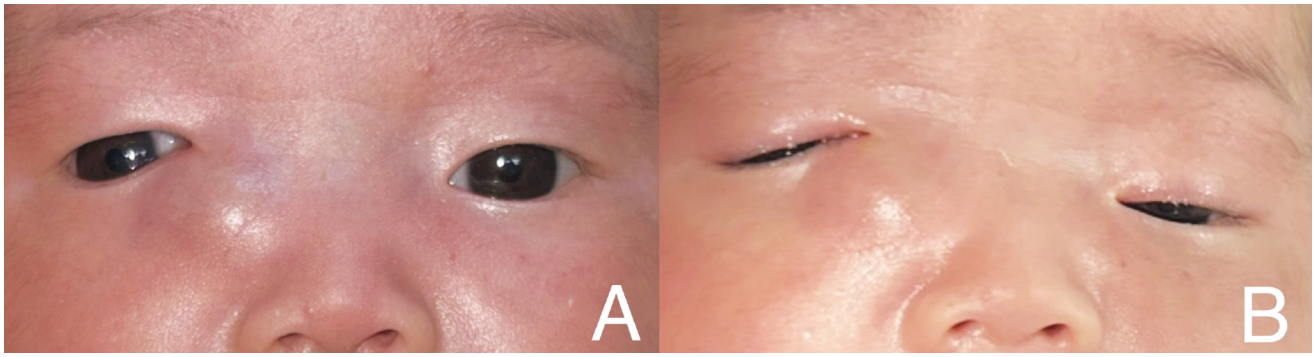


Figure 3 Clinical appearance at presentation to Songklanagarind Hospital. (A) Enlarged right dacryocystocele causing superotemporal displacement of the right globe. (B) After 2 days of intravenous ceftriaxone therapy, showing improvement in inflammation, although the enlarged dacryocystocele and globe displacement persist.

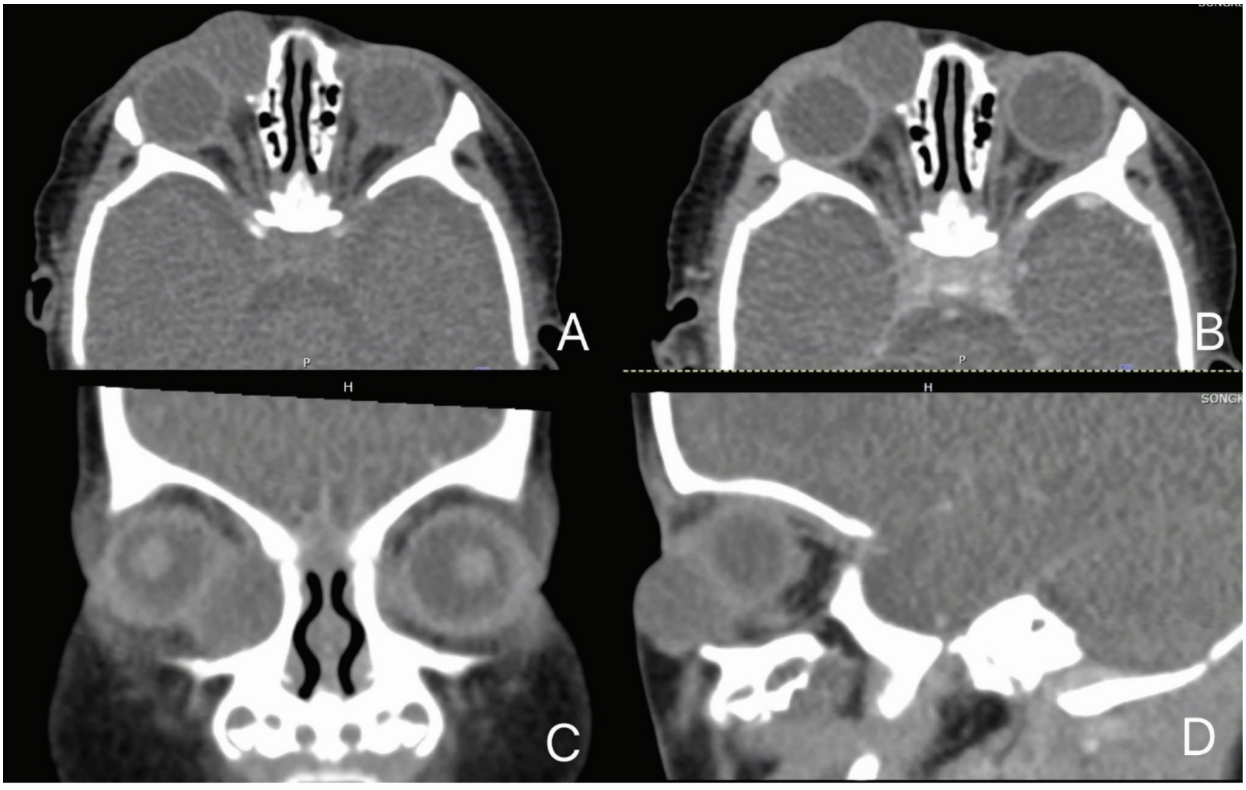


Figure 4 Orbital computed tomography (CT) findings. (A) Non-contrast axial CT demonstrates a large, hypodense, cystic lesion at the right medial canthus. (B) Contrast-enhanced axial CT shows subtle peripheral rim enhancement without internal enhancement. (C) Coronal CT demonstrates superotemporal displacement of the right globe secondary to orbital mass effect. (D) Sagittal CT demonstrates inferior compression of the globe with distortion of its contour caused by the enlarged dacryocystocele.

Nasolacrimal duct probing of the right eye was performed at 21 days of age. Intraoperatively, compression of the lacrimal sac expressed purulent discharge through both the upper and lower puncta. The probing was completed successfully without complications. The globe returned to its normal position immediately after decompression. At follow-up, the medial canthal swelling had completely resolved, with restoration of the normal globe position and no evidence of recurrence to date (Figure 5). Culture of the purulent discharge grew Enterococcus faecalis and Staphylococcus aureus. Following clinical improvement after probing, intravenous cefazolin was continued for an additional 5 days before being transitioned to oral cephalexin.

Written informed consent for publication was obtained from the patient's legal guardian.



Figure 5 Clinical outcome following nasolacrimal duct probing. (A) Postoperative day 1, showing marked reduction in the dacryocystocele with improvement in globe position. (B) Postoperative day 2, demonstrating further resolution of the lesion and restoration of normal globe alignment. (C) Two weeks postoperatively, showing complete resolution of the dacryocystocele without recurrence.

Discussion

Congenital dacryocystocele is an uncommon congenital anomaly of the lacrimal drainage system, accounting for approximately 0.1%–0.3% of cases of congenital nasolacrimal duct obstruction.[1,2] It is thought to result from simultaneous proximal obstruction at the valve of Rosenmüller and distal obstruction at the valve of Hasner, leading to fluid accumulation and cystic distension of the lacrimal sac.[3,4] Previous studies have demonstrated a marked female predominance (approximately 73%) and predominantly unilateral involvement (approximately 88%).[1,4]

Obstruction of the lacrimal drainage system results in the accumulation of fluid within the lacrimal sac, which initially contains mucoid material and presents clinically as a tense, grayish-blue cystic swelling inferior to the medial canthus.[3,5] The cyst contents may include mucus, amniotic fluid, tear fluid, and cellular debris, while secondary bacterial colonization can lead to progressive enlargement and subsequent infection.[3] Diagnosis is typically based on the characteristic appearance of a bluish cystic mass in the medial canthal region, often accompanied by epiphora since birth and recurrent conjunctivitis.[5,6] Although some congenital dacryocystoceles resolve with conservative management, secondary acute dacryocystitis may develop within days to weeks, necessitating systemic antibiotic therapy. Persistent or complicated cases frequently require surgical intervention to relieve the obstruction and prevent further complications.[3,6]

Ultrasonography is the preferred initial imaging modality because it is non-invasive and readily distinguishes cystic from solid or vascular lesions. Reported dacryocystocele sizes on ultrasonography range from 9.6 to 16.3 mm, with a mean diameter of 11.5 mm.[3] In the present case, however, orbital CT was performed because of the unusual finding of globe displacement. CT demonstrated a large cystic lesion measuring 17.7 × 14.6 × 16.0 mm arising from the lacrimal sac, producing significant orbital mass effect with superotemporal displacement of the globe. Importantly, there was no extension into the nasal cavity or evidence of respiratory compromise.

To the best of our knowledge, mechanical displacement of the entire globe secondary to congenital dacryocystocele has not previously been reported. Although ocular complications of dacryocystocele resulting from external compression are uncommon, compression-induced astigmatism has been described. Shekunov et al. reported induced astigmatism in 1 of 9 patients (11%) with congenital dacryocystocele, while Bogan et al. found astigmatism greater than +1.25 diopters, reaching as high as +5.50 diopters, in 8 of 12 affected eyes.1,6 These observations suggest

that a sufficiently large dacryocystocele can exert mechanical pressure on the globe and alter ocular anatomy. In contrast, the mass effect in our patient was considerably greater, resulting in displacement of the entire globe rather than corneal deformation alone. This case therefore expands the spectrum of compression-related ophthalmic manifestations of congenital dacryocystocele and demonstrates that giant lesions may behave as orbital masses.

Congenital dacryocystoceles complicated by acute dacryocystitis are best managed with systemic antibiotics and early surgical intervention.2 Empirical antibiotic therapy should provide adequate coverage against the most common causative organisms, particularly MSSA and Streptococcus species. In our patient, antibiotic therapy was changed from ceftriaxone to cefazolin because cefazolin provides excellent coverage against these pathogens and has more reliable activity against MSSA than ceftriaxone. Furthermore, cefazolin is generally preferred in neonates because ceftriaxone may displace bilirubin from albumin-binding sites, increasing the risk of kernicterus, and may form calcium-ceftriaxone precipitates when administered concomitantly with calcium-containing solutions. Nasolacrimal duct probing has been shown to be effective for complicated dacryocystoceles, including those associated with dacryocystitis or preseptal cellulitis, often resulting in complete resolution without the need for silicone intubation.[7,8] In our patient, probing resulted in immediate decompression of the dacryocystocele, resolution of the obstruction, and restoration of the globe to its normal position, confirming that the globe displacement was caused by mechanical compression from the enlarged lacrimal sac.

Microbiological culture yielded *Staphylococcus aureus* and *Enterococcus faecalis*. *Staphylococcus aureus* is a well-recognized pathogen in acute dacryocystitis and was likely the primary causative organism in this case. Although *E. faecalis* was also isolated, its clinical significance is uncertain and may represent colonization or contamination. Nevertheless, the patient's favorable response to intravenous cefazolin therapy combined with surgical drainage, with no recurrence during follow-up, supports the effectiveness of this management strategy.

This case highlights a rare and previously unreported presentation of giant congenital dacryocystocele causing significant orbital mass effect with globe displacement. Clinicians should recognize that, although congenital dacryocystoceles typically present as medial canthal cystic masses, exceptionally large lesions may produce orbital signs that mimic primary orbital pathology. Early recognition, appropriate imaging when atypical features are present, and timely surgical intervention are essential to prevent potentially vision-threatening and systemic complications while achieving excellent anatomical and functional outcomes.

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ภาวะถุงน้ำตาโป่งพองจากท่อน้ำตาอุดตันแต่กำเนิดขนาดใหญ่ทำให้เกิดการกดเบียดและเคลื่อนที่ของลูกตาในทารก



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บทคัดย่อ:

บทนำ: ภาวะถุงน้ำตาโป่งพองจากท่อน้ำตาอุดตันแต่กำเนิด (congenital dacryocystocele) เป็นภาวะที่พบไม่บ่อย และจากการทบทวนวรรณกรรมของผู้เขียน ยังไม่พบการรายงานของภาวะนี้ที่ก่อให้เกิดการกดเบียดลูกตาจนทำให้ลูกตามีการเคลื่อนที่

รายงานผู้ป่วย: ผู้ป่วยทารกเพศหญิงอายุ 19 วัน มีก้อนบวมสีคล้ำบริเวณหัวตาด้านขวาตั้งแต่แรกเกิด ก้อนมีขนาดใหญ่ขึ้นและมีการอักเสบติดเชื้อของถุงน้ำตา (dacryocystitis) แทรกซ้อน หลังได้รับยาปฏิชีวนะทางเส้นเลือด พบว่าการอักเสบลดลง แต่ก้อนยังมีขนาดใหญ่และดันลูกตาไปทางด้านบนและด้านข้าง การตรวจเอกซเรย์คอมพิวเตอร์พบก้อนลักษณะเป็นถุงน้ำ ขนาด 17.7 × 14.6 × 16.0 มม. บริเวณมุมตาด้านใน ต่อเนื่องกับท่อน้ำตาในจมูก และก่อให้เกิดแรงกดของก้อนต่อลูกตา ผู้ป่วยได้รับการรักษาต่อด้วยยาปฏิชีวนะทางเส้นเลือดร่วมกับการแยงท่อน้ำตา ส่งผลให้ก้อนยุบลงทันที และลูกตากลับสู่ตำแหน่งปกติ

สรุป: ภาวะถุงน้ำตาโป่งพองจากท่อน้ำตาอุดตันแต่กำเนิดขนาดใหญ่สามารถก่อให้เกิดแรงกดต่อลูกตา และทำให้ลูกตาเคลื่อนตำแหน่งได้ การวินิจฉัยและการรักษาอย่างทันท่วงทีมีความสำคัญ เพื่อป้องกันภาวะแทรกซ้อนรุนแรงต่าง ๆ ที่อาจเกิดขึ้นได้

คำสำคัญ: ภาวะถุงน้ำตาโป่งพองจากท่อน้ำตาอุดตันแต่กำเนิด, การอักเสบบริเวณถุงน้ำตา, ลูกตาเคลื่อนตำแหน่ง, ท่อน้ำตาอุดตันแต่กำเนิด, การแยงท่อน้ำตา

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