

## Case Report

# Bilateral Congenital Dacryocystocoeles with Concurrent Intranasal Mucocoeles Causing Respiratory Distress in a Neonate

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Congenital dacryocystocoeles are uncommon, presenting as a fluctuant bluish mass inferior to the medial canthus. Even more rarely, these dacryocystocoeles are complicated by intranasal extension. We present a case of a newborn infant with bilateral dacryocystocoeles with intranasal extension (intranasal mucocoeles) causing respiratory distress and feeding difficulties. Prompt surgical correction was performed, with the mucocoeles being de-roofed, leading to the resolution of the dacryocystocoeles. The aetiology, clinical features, and therapeutic options are discussed. [*Asian J Surg* 2006;29(2):109-11]

**Key Words:** dacryocystocoele, nasal mucocoele, nasolacrimal duct cyst, neonatal respiratory distress

## Introduction

A congenital dacryocystocoele is an uncommon disease entity that is usually treated by ophthalmologists. When uncomplicated, they may be successfully treated conservatively. Rarely, these dacryocystocoeles can be complicated by intranasal extension presenting as a mucocoele, causing respiratory distress or feeding difficulties in the newborn, especially when bilateral. When this occurs, an otolaryngologist will be involved for prompt investigation and surgical intervention. As such, an intranasal mucocoele must be considered as a differential diagnosis when assessing neonates with respiratory distress.

## Case report

A 1-day-old female neonate was noted to have a pale bluish cystic mass overlying the left nasolacrimal duct. She was born at term, of normal birth weight, following a normal vaginal delivery. Her Apgar scores were 7 and 9 at 1 and 5 minutes, respectively. She had mild respiratory distress on day 1, and her paediatrician noted the cystic mass. She was

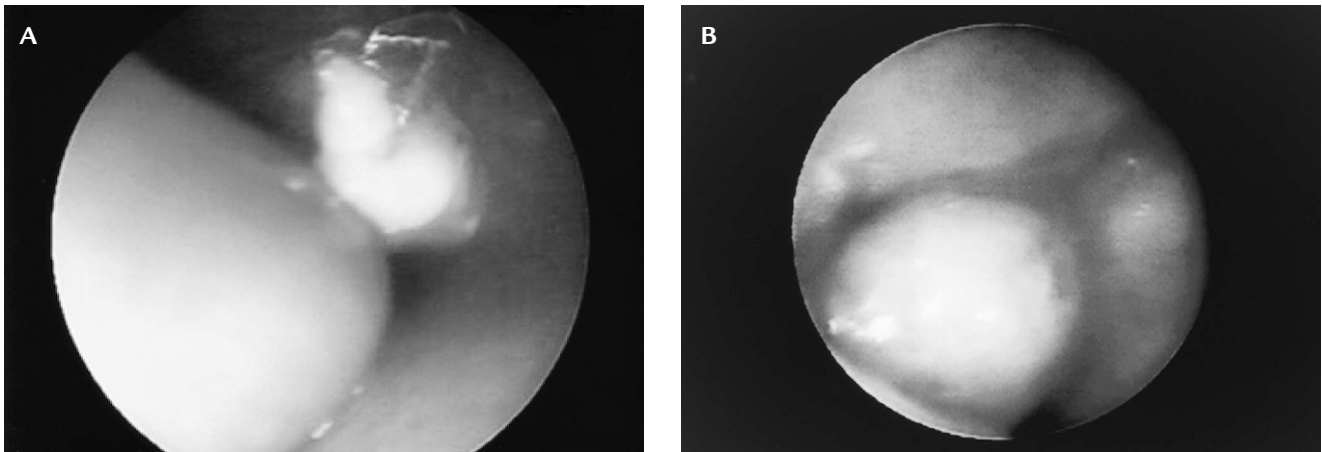
initially treated with topical chloramphenicol ointment to her left eye and local compressive massage.

Otolaryngological consultation was requested regarding nasal congestion and intermittent respiratory distress with accompanying mild oxygen desaturation. Poor feeding secondary to nasal congestion was also of concern, and the infant was requiring nasogastric feeding. No other significant features in the history were elucidated, except that the elder brother of the patient had a similar unilateral dacryocystocoele noted shortly after birth that resolved spontaneously.

On examination, bilateral pale bluish fluctuant masses at the medial canthus were noted, the left larger than the right, giving the appearance of apparent hypertelorism. There was mucoïd accumulation on the conjunctiva on both sides. Examination of the nasal cavities revealed bilateral, smooth, mucosa-covered, cystic swellings protruding from below the inferior turbinate (Figure 1), more apparent on the left side. These cystic masses were causing near complete obstruction on the left and restriction of airflow on the right. There was also an accumulation of nasal mucus compounding the obstruction on the left.

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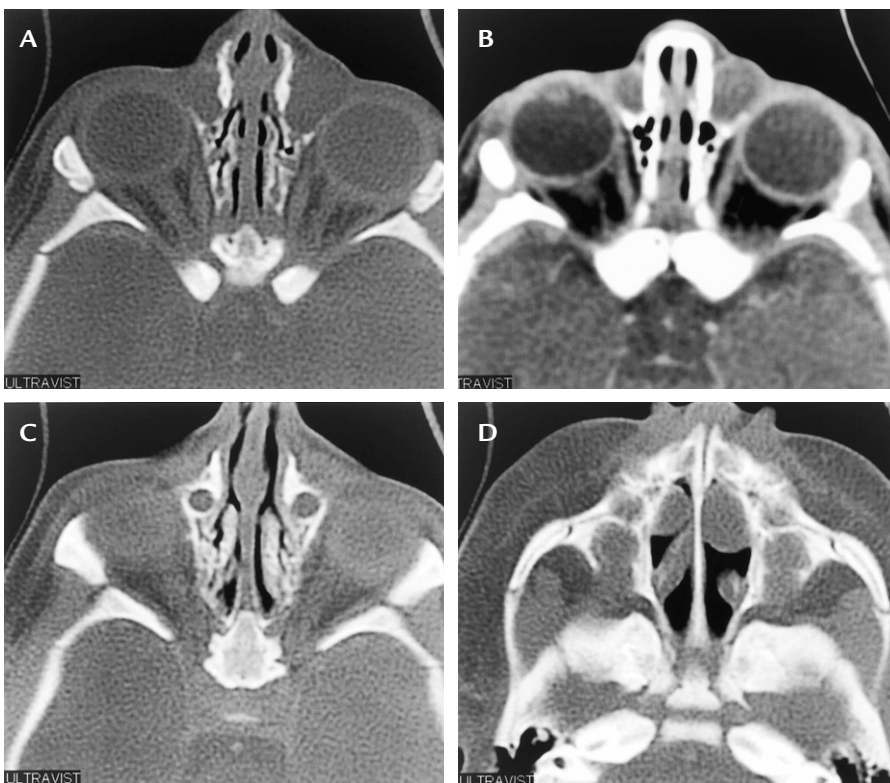
**Figure 1.** Endoscopic view of the nasal cavities: (A) incomplete obstruction with mucopus, possibly secondary to stasis, in the right nostril; (B) near complete obstruction due to a mucocoele in the left nostril.

The rest of the otolaryngological examination was normal, and there were no features of syndromic or non-syndromic congenital abnormalities.

A computed tomography (CT) scan showed low density, cystic lesions in the region of the inferior meatus bilaterally, in the location of the distal nasolacrimal duct (Figure 2). The nasolacrimal ducts were found to be expanded bilaterally, most marked on the left, also causing widening of the proximal duct. The nasal choanae were found to be patent, and there was narrowing and medial deviation of the posterior

nasal wall, consistent with choanal stenosis. No further abnormalities were identified on CT.

Two days after conservative management was initiated, surgical correction was performed as there was no improvement in the patient's condition. Intraoperatively, pale blue cystic lesions in the inferior meatus were found. These mucocoeles were endoscopically de-roofed, expelling mucopurulent fluid. The dacryocystocoeles resolved immediately following this procedure. The nasolacrimal ducts were then probed, and irrigated bilaterally, with free flow of saline. Post-



**Figure 2.** Series of axial computed tomography scans. (A) Without contrast: dilated nasolacrimal sac. (B) With contrast: dilated nasolacrimal sac. (C) Dilated nasolacrimal ducts. (D) Bilateral nasal mucocoeles.

operative recovery was uneventful and the patient was discharged home on the second postoperative day.

## Discussion

Impatency of the nasolacrimal duct at birth is quite common. It is generally reported that approximately 30% are closed at birth, although impatency has been reported to be as high as 84% and as low as 6%.<sup>1,2</sup> In one study, approximately 75% of fetuses that underwent autopsy had a membranous obstruction of the nasolacrimal system.<sup>3</sup> Despite impatency being quite common, only 2–6% of newborn children are symptomatic.<sup>2,3</sup> The dacryocystocoele is a dilation of the lacrimal sac that occurs with obstruction of the proximal and distal parts of the lacrimal drainage system. In congenital forms, a dacryocystocoele is formed by obstruction of the valve of Hasner (the opening beneath the inferior turbinate) with coexistent obstruction of the valve of Rosenmuller (the junction of the common canaliculus and the lacrimal sac). Some authors believe that the valve of Rosenmuller acts as a one-way valve to allow fluid in, but not out.<sup>2</sup> Equivalent nomenclature for dacryocystocoele includes amniotocoele, amniocoele, lacrimal sac mucocoele and dacryocoele.

The incidence of dacryocystocoeles is three to four times higher in female infants than male, attributed to a narrower nasolacrimal duct in females.<sup>4</sup> A dacryocystocoele presents as a fluctuant bluish mass inferior to the medial canthus causing epiphoria and accumulation of mucopurulent secretions. If infected, the patient may have facial cellulitis. Dacryocystocoeles are bilateral in about 14% of cases.<sup>1,5</sup> If the dacryocystocoele is associated with an intranasal mucocoele, as in our case, a cystic mass can be seen within the nasal cavity. The incidence of a dacryocystocoele with concurrent intranasal mucocoele is unknown, with only a few cases reported in the literature.<sup>6</sup> This can lead to respiratory distress in a newborn, who is an obligate nasal breather, if the involvement is bilateral.<sup>4</sup>

The differential diagnosis of dacryocystocoeles with intranasal extension includes other paranasal masses such as nasal encephalocoele, dermoid, haemangioma, nasal glioma and dacryocystitis. If a nasal mass is not visualized in a neonate with nasal obstruction, with or without respiratory distress, neonatal rhinitis and choanal atresia must be included in the differential diagnosis.<sup>4</sup>

Radiological imaging is important in assessing the extent of the mass, and is essential in ruling out any intracranial communication prior to surgical intervention. A CT scan is

the first-line imaging modality for a dacryocystocoele with intranasal extension, though dacryocystography, ultrasonography and magnetic resonance imaging have been used. A CT scan will demonstrate a medial canthal mass, a dilated ipsilateral nasolacrimal duct, and an intranasal cystic mass.

In the vast majority of neonates with symptomatic nasolacrimal sac obstruction, the obstruction will spontaneously resolve by the time the infant is 6 months of age, with 85% being reportedly open at 1 year.<sup>7</sup> As such, the management of uncomplicated nasolacrimal sac obstruction is conservative for the first 4–6 months of life or more, consisting of lacrimal sac massage, warm compress, or if complicated by infection, the addition of topical antibiotics. Lacrimal probing may be required if spontaneous resolution does not occur, or if complications such as severe or recurrent dacryocystitis, corneal astigmatism with the threat of anisometric amblyopia, or permanent canthal asymmetry occur.<sup>4</sup> When nasolacrimal duct obstruction is complicated by an intranasal mucocoele with respiratory distress, prompt surgical treatment is mandatory, with marsupialization of the mucocoele the treatment of choice.

In conclusion, intranasal extension of dacryocystocoeles should be considered when assessing a neonate with respiratory distress or feeding difficulties. CT scanning to rule out intracranial communication should be performed after nasal examination, and prior to surgical correction. If symptomatic nasal obstruction is present, prompt surgical treatment is mandatory.

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