Bilateral dacryocystocele with an intranasal cyst as the cause of respiratory distress in a newborn

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Abstract. Bilateral dacryocystocele with an intranasal cyst as the cause of respiratory distress in a newborn. A congenital dacryocystocele with an intranasal cyst is an uncommon lesion that is usually treated by ophthalmologists, although sometimes an otolaryngologist is consulted first because of nasal obstruction. The nasal cavity is narrow in newborns and can easily be obstructed, even by small lesions. Prolapse or expansion of the cyst into the nose may lead to respiratory distress and difficulty in feeding, since newborns are obligate nose breathers. Here we report a case of bilateral dacryocystocele with intranasal extension in a 3-day-old female infant. The infant presented with respiratory distress and episodic desaturation and was managed successfully by bilateral endoscopic marsupialization of the intranasal cysts. This case report discusses the diagnosis and management and reviews the relevant literature. These findings suggest that congenital dacryocystocele with an intranasal cyst must be considered in the differential diagnosis of newborns suffering from nasal respiratory difficulty.

Introduction

Obstruction of the nasolacrimal duct (ND) is a common congenital abnormality that is reported in up to 75% of all newborns.1 In most cases, the obstruction is asymptomatic and resolves spontaneously within the first year of life.2 A symptomatic obstruction can present as a cystic mass at the medial canthal region or in the nasal cavity.1 Due to the presence of eye symptoms in the majority of patients, congenital dacryocystoceles with or without intranasal cysts are usually diagnosed and treated by ophthalmologists. An otorhinolaryngologist may be the first specialist to be consulted when isolated ND cysts are not visible externally and do not cause ophthalmologic symptoms.3

The nasal airway can easily be obstructed, even with small lesions, since the nasal cavity is narrow in newborns. Prolapse or expansion of the cyst into the nose may lead to respiratory distress and difficulty in feeding, as newborns are obligate nose breathers. In addition to respiratory distress, which can threaten the infant’s life, dacryocytis can occur, which may lead to preseptal cellulitis.3,5 Prenatal or early diagnosis is very important in preventing these complications, which are responsible for morbidity and mortality.

Case report

A 3-day-old female infant in the neonatal intensive care unit was referred for an otolaryngological examination for feeding difficulty and for respiratory distress that usually improved with crying. The patient also had mucopurulent ocular discharge from the left inferior punctum (Figure 1A). Endoscopic nasal examination of the patient, who had respiratory distress and intercostal retractions, revealed bilateral soft tissue lesions arising from the inferior meatus that were occluding the nostrils (Figure 1B,C). Magnetic resonance imaging (MRI) revealed cystic dilatation of the both left and right lacrimal sacs and ND and ND cysts at the both

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intubation was performed bilaterally to confirm the patency of the nasolacrimal duct and to prevent recurrence (Figure 2C,D). The patient’s respiratory distress improved, and the symptoms of infection returned.

inferior meatus (Figure 1D,E). After the purulent cyst content was drained, endonasal endoscopic marsupialization was performed by removing the medial wall of the cyst (Figure 2A,B). Silicone tube intubation was performed bilaterally to confirm the patency of the nasolacrimal duct and to prevent recurrence (Figure 2C,D). The patient’s respiratory distress improved, and the symptoms of infection returned.
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resolved immediately after the operation. Endoscopic examination at the third postoperative month revealed full epithelialization of the marsupialised cyst regions in the inferior meatus and good tear drainage (Figure 2E,F). There was no recurrence during the one-year follow-up period.

Figure 2
(A, B) After the content of the purulent cyst was drained, the medial wall of the cyst was marsupialised. (C, D) Silicone tubes were inserted bilaterally into the nasolacrimal drainage system. Examination at the third postoperative month revealed epithelialization of the marsupialised cyst on the (E) right and the (F) left sides.
Discussion

Distal obstruction of the nasolacrimal duct is a common condition in neonates that results in epiphora, an overflow of tears and a mucoid ocular discharge. It usually resolves spontaneously, but obstruction results in the development of an intranasal lacrimal duct cyst or dacryocystocele in 2-6% of children with a non-patent nasolacrimal duct.6,9 There may be an inherited predisposition to dacryocystocele formation as there is an increased prevalence in females and an increased incidence in Caucasian and Hispanic populations.7,2,10,11

In newborns, the canalization of the lacrimal duct is often not fully developed. Thus, there is often connate obstruction of the lacrimal ducts, and about 75% of all newborns have a persisting Hasner’s membrane at the opening of the nasolacrimal duct into the lower nasal meatus.7 However, there are symptoms of nasolacrimal duct obstruction and dacryocystocele in only 25 neonates per 100,000 live births.2 Although dacryocystocele is considered a neonatal disease, it has also been reported in adults.12

When the Hasner valve is imperforate, the lacrimal sac fills and becomes distended with amniotic fluid and mucous secretions. This cystic distension of the lacrimal sac creates the dacryocystocele that compresses the canal system. This causes a trapdoor-type blockage and functional or mechanical proximal obstruction at the junction of the inferior canaliculi and lacrimal sac, which is called the Rosenmüller valve. Furthermore, the cystic expansion of the nasolacrimal duct mucosa into the nose can form an intranasal cyst.5,8 This cystic lesion can protrude into the nasal lumen, causing nasal obstruction and respiratory distress. The level of the distal obstruction can indicate whether the dacryocystocele is a mass at the medial canthus and/or is an intranasal cyst.2,3,13 Since a cystic mass in the medial canthal region is the main clinical finding in dacryocystocele cases, the differential diagnosis should consider both benign and malignant lesions, such as hemangioma, lymphangioma, teratoma and rhabdomyosarcoma, and developmental lesions, such as nasal glioma, encephalocele, dermoid cyst and nasolabial cyst.5,2,14

In 20%-70% of dacryocystocele cases, the dacryocystocele is accompanied by intranasal cysts, and neonatal respiratory distress may occur, especially in bilateral cases.7,1,12 But as shown by this report, an intranasal cyst can exist as an isolated nasal cyst detected in the inferior meatus without the coexistence of a medial canthal mass or eye symptoms.7 In the present case, the dacryocystocele was clinically invisible, but MRI showed a bilaterally enlarged lacrimal sac that was consistent with a dacryocystocele. Since the nasal cavity in newborns is narrow and newborns are obligate nasal breathers during their first three weeks of life, bilateral nasal obstruction can easily lead to respiratory distress. A differential diagnosis of dacryocystocele with an intranasal cyst that causes nasal obstruction and respiratory distress should consider other causes of nasal obstructions, such as choanal atresia and stenosis, congenital deformities and cysts, congenital neoplasms and inflammation, and cysts secondary to infection.2,4,13

Performing ultrasonography and MRI during the 3rd trimester can result in prenatal diagnosis of a dacryocystocele, which is important to prevent possible morbidity and mortality.2,6 However, a definitive diagnosis is made by bilateral nasal endoscopic examination during the neonatal period. Respiratory distress that appears during feeding and improves while crying first brings to mind choanal atresia. To confirm the diagnosis, the examiner should use a nelaton catheter: the catheter will pass along the blocked nasal passage because of the cyst’s elasticity, causing the nasal passage to appear to be open. If nasal endoscopy is not part of the physical examination, it is impossible to correctly evaluate the inferior meatus, and nasolacrimal duct cysts will be missed.

Radiological imaging is recommended to clarify the diagnosis and to determine the extent of the cyst. Dacryocystography, ultrasonography, MRI and CT scanning are all techniques that can be helpful.

Treatment recommendations for dacryocystocele with and without intranasal mucoceles vary widely in the literature. The suggested treatments include warm compresses and lacrimal sac massage, topical and/or intravenous antibiotics, surgical probing of the nasolacrimal duct with or without silicone tube intubation and marsupialization of the intranasal mucocele.2,3,12 Initially, a conservative approach (i.e., warm compresses, massage) is recommended. If it is not resolved, probing may be the next choice. Although resolution of the nasal cysts is possible with supportive management (in about 30% of cases), surgical treatment is usually needed due to
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complications, which can include dacryocystitis, cellulitis, astigmatism and narrowing of the lid fissure due to a large cyst, respiratory difficulty due to a nasal cyst and non-resolution of the cyst after a short trial period of massage.⁴,⁷,¹³-¹⁵

Although our patient had a purulent secretion from the left inferior punctum, surgery was required because of her respiratory distress. During surgery, nasal cysts can be marsupialized with aids such as forceps, power instruments, lasers, etc.³,⁴,⁹ Some physicians insert lacrimal probes through the nasolacrimal duct and rinse the lacrimal ducts with normal saline solution following the marsupialization to confirm the patency of the nasolacrimal duct and to exclude proximal obstructions.⁴,⁸,¹³ Others use prolonged silicone intubation after marsupialization of the cyst to confirm the patency of the nasolacrimal duct and to prevent recurrence.²,³

Conclusion

Dacryocystocele with an intranasal cyst should be considered in the differential diagnosis of newborns with nasal respiratory difficulty. Endoscopic examination plays an important role in the diagnosis and must be performed during the routine physical examination for evaluating respiratory distress in newborns. When emergency treatment is required for complicated dacryocystoceles, the most common treatment option is endoscopic endonasal marsupialization of the cyst walls.

References