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Case report

Congenital bilateral dacryocystocele

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ABSTRACT

Introduction: Newborns are obligatory nasal breathers. Therefore, nasal obstruction can lead to cyanosis and desaturation. In spite of being very rare, congenital bilateral dacryocystocele is a possible etiology for neonatal respiratory distress.
Case summary: Case report of a male newborn with respiratory distress caused by a bilateral polypoid and bluish lesion occupying almost the entire inferior nasal meatus. Imaging confirmed bilateral dacryocystocele. Treatment was conservative. There was spontaneous drainage, with relief of respiratory distress. Discussion The diagnosis of congenital dacryocystocele is clinical, although imaging exams may be requested to confirm it. Treatment is controversial, because the natural history is variable. An initial conservative management may be recommended, but, if there is a permanent respiratory obstruction without improvement, surgical management is mandatory.

1. Introduction

Newborns have an obligatory and exclusive nasal breathing. Therefore, an obstruction at this level can lead to respiratory distress, demanding an orientation plan for treatment in urgent care setting [1].

In spite of not being a nasal disease, laryngomalacia is the most frequent cause of neonatal respiratory obstruction [1]. Meanwhile, choanal atresia is a well-defined etiology of neonatal upper respiratory obstruction [2], being one of the most common nasal congenital anomalies. In almost 50% of the cases, it is associated with other congenital alterations, such as the signs that define CHARGE syndrome [3].

Meningocele/meningoencephalocele and dacryocystocele (nasolacrimal duct cyst) may also be responsible for congenital nasal obstruction. When a rhinoscopy is performed, both appear as cystic or polypoid mass lesions occupying a substantial portion of the nasal cavity [4,5]. Congenital dacryocystocele is very rare, being observed in only 0.1% of newborns with nasolacrimal obstruction [1]. There is bilateral lesion in up to 25% of these cases [6].

2. Case report

The patient was a two-day-old male newborn, who was dependent on high-flow nasal cannula oxygen and orogastric feeding tube and presented respiratory distress (frequent episodes of cyanosis and desaturation). In ENT physical examination, the patient showed nose flaring, accessory muscle use and hypertelorism (Fig. 1). Nasal endoscopy revealed a bilateral polypoid lesion with bluish colored walls, occupying almost the entire inferior nasal meatus.

Computed tomography (CT) and magnetic resonance imaging (MRI) (Fig. 2) confirmed the diagnosis of bilateral dacryocystocele.

Fig. 1. Hypertelorism before the beginning of treatment.

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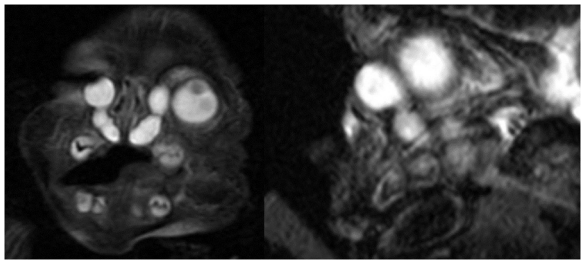

Fig. 2. Magnetic resonance imaging (MRI) confirming the presence of bilateral dacryocystocele.

After conjunct observation with ophthalmology, it was decided to start treatment in a conservative way: lacrimal sac massage and topical ophthalmic antibiotherapy during one week. There was spontaneous drainage, with progressive relief of respiratory distress and disappearance of hypertelorism. After one, six and twelve months, nasal endoscopy (Figs. 3 and 4), and ophthalmological examination were normal. The patient's facies became normal as well (Fig. 5).

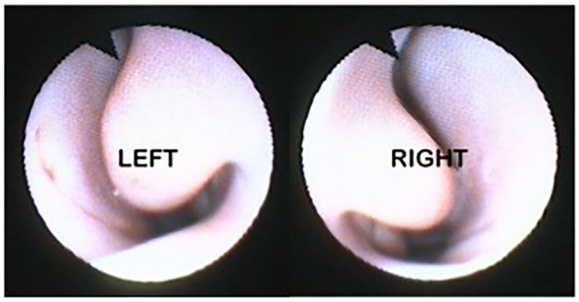

Fig. 3. Normal bilateral nasal endoscopy, at one-month follow-up.

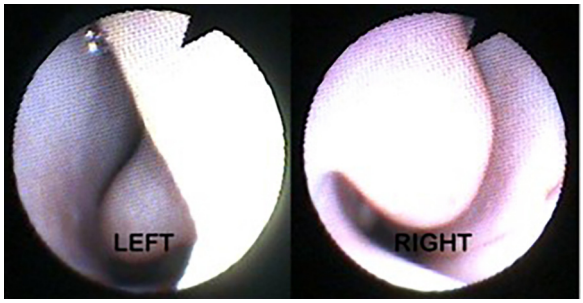

Fig. 4. Normal bilateral nasal endoscopy, at twelve-month follow-up.

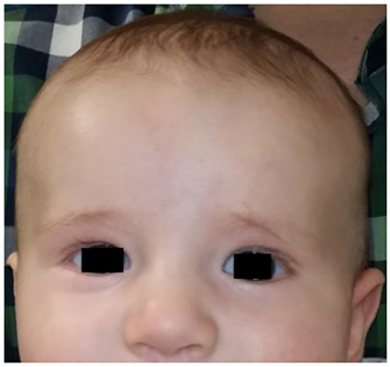

Fig. 5. Normal facies at twelve-month follow-up.

3. Discussion

Dacryocystocele results from the obstruction of two levels of lacrimal system: valves of Rosenmuller (proximal) and Hasner (distal) [5]. The valve of Hasner is a fold of mucous membrane, which communicates the nasolacrimal duct with the inferior nasal meatus and is supposed to be completely perforated after the sixth month of fetal development [7]. However, if the membrane is unperforated, lacrimal secretion persists, but it is not drained into the nasal cavity [6]. The swollen sac and nasolacrimal duct lead to functional sac-canalicular junction obstruction at the valve of Rosenmuller [8].

The diagnosis is clinical, with a very important role of the otorhinolaryngologist. CT and MRI are also very useful, having similar sensitivity to identify this lesion [9].

Since the natural history is variable, there is no universal therapeutic strategy [5]. Nevertheless, an initial conservative management with warm compresses and external lacrimal sac massage may be recommended [10]. If there is a permanent respiratory obstruction without improvement, a surgical treatment is mandatory. It consists in a combined technique, using a lacrimal probe and distal endoscopic marsupialization of the cystic sac, in order to prevent recurrence [7,8].

In this clinical case, there was a bilateral lesion resulting in respiratory distress. However, the absence of dacryocystis and the immediate response to conservative management avoided surgery.

4. Conclusion

Nasal endoscopy and imaging have a crucial role in the diagnosis of neonatal respiratory distress causes. Otorhinolaryngological intervention is also critical in the surgical management. Therefore, otorhinolaryngologists should be very exhaustive in the work-up of these cases.

Disclosure of interest

The authors declare that they have no competing interest.

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