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Case Report

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Bilateral Nasolacrimal Duct Congenital Mucocele in a Newborn

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Abstract

A new-born at 38+0 weeks of gestational age presented with respiratory distress (resolved with oxygen administration) and swelling along the inner corner of the left eye with a deformed profile of the palpebral fissure in the medial canthal region. CT scan showed a hypo dense formation with fluid/suprafluid density, originating from the left nasolacrimal duct and extended caudally into the inferior nasal meatus and a smaller one, the right nasal inferior meatus. Histologic examination showed the lesion to be compatible with nasolacrimal mucoceles.

Keywords: Nasolacrimal Duct; Dacryocystocele; Dacryocystitis; Nasolacrimal Duct Obstruction Endoscopic Treatment.

Introduction

A congenital mucocele of the nasolacrimal duct is an uncommon lesion that results from the mal-development of the nasolacrimal drainage system [1-5]. Complications of this condition can include epiphora, dacryocystitis, cellulitis, sepsis, and respiratory distress [2]. Computed tomography (CT) has been shown to play an essential role in diagnosing this entity [1]. Surgical treatment with marsupialisation of the mucocele in new-borns is rarely described and prescribed in case of nasal obstruction that does not resolve spontaneously with conservative treatment.

Case Report

From a normal pregnancy, a female new-born born at 38+0 weeks of gestational age and birth weight 3295 grams by elective caesarean section to a second pregnancy 0 para mother (1 previous caesarean section), Apgar score 9-9, showed at 15 minutes of life respiratory distress requiring oxygen administration. For this reason, the new-born was admitted to the Neonatal Intensive

Care Unit (NICU), where swelling was observed along the left internal medial canthus, which deformed the profile of the eyelids (Figure 1). During her stay in the NICU, the respiratory symptoms resolved rapidly (FiO2 of 0.25 for 24 hours), so after one day the new-born was re-united with her mother in the Rooming-In department: she was eupnoic with normal values of SpO2 (98%) and heart rate (120 bpm), good air penetration bilaterally in the thorax, abdomen not painful on superficial or deep palpation. Given the suspicion of mucocele of the lacrimal ducts at the ophthalmological examination, a conservative treatment was performed with massage of the external lacrimal sac, antibiotic eye drops and a CT scan was requested. The ENT physical examination showed a greyish cystic lesion in the left nasal cavity under the inferior turbinate that appeared cranially displaced. The new-born girl presented with a cystic-looking swelling in the left medial canthus region and in the upper eyelid (Figure 1). A small round cystic reddish lesion was observed in the inferior meatus below the inferior turbinate in the right nasal fossa. The use of endoscopy in the operating room allowed the exploration of both nasal passages to exclude a small dacryocystocele which supported the radiological examination carried out. A CT scan of the paranasal sinuses without contrast documented the presence of a hypo dense formation with fluid/suprafluid density, with a total axial diameter of approximately 22 mm, originating from the left nasolacrimal duct and extending caudally into the nasal meatus ipsilateral inferior in an hourglass shape. This lesion cranially appeared as a hypo dense mass localized in the left medial orbital canthus (widening of the lacrimal sac). This hypo dense formation was compatible in the first hypothesis with the mucocele of the left nasolacrimal duct. On the right side, another similar, smaller lesion was evident in the nasal meatus inferior (Figure 2). Given the non-resolution with conservative treatment after ten days and the high risk of systemic infection, the patient underwent surgical therapy to drain and marsupialize the dacryocele. Under general anaesthesia, a dacryocystocele with an external component and an endonasal component under the inferior turbinate was appreciated during endoscopic surgical procedures supported by the figure of the ophthalmologist for combined approach. After probing and cannulating the inferior lacrimal canaliculus, the external component was drained, and the cyst was marsupialized at the level of the inferior meatus until it was highlighted and freed the opening of the nasolacrimal duct. Also, the right nasal cavity was explored, and a cyst was seen at the level of the inferior meatus below the head of the inferior turbinate, which was marsupialized, resulting in the leakage of purulent material. The inferior lacrimal canaliculus was probed and cannulated, and the probe was observed at the level of the inferior meatus (Figure 3). Histologic examination of the nasal component bilaterally confirmed the wall of a normal structured lacrimal sac, normotypical with notes of fibrosis of the chorion; the finding was compatible with dacryocystocele. Ten days following surgery (Figure 4), the patient was doing well; there was complete resolution of abnormalities, there were no signs of dacryocystitis bilaterally, no pathological secretions were observed when squeezing the lacrimal caruncle, patent nasal passages, no stagnation of serum-mucous secretions in the nasal passages.

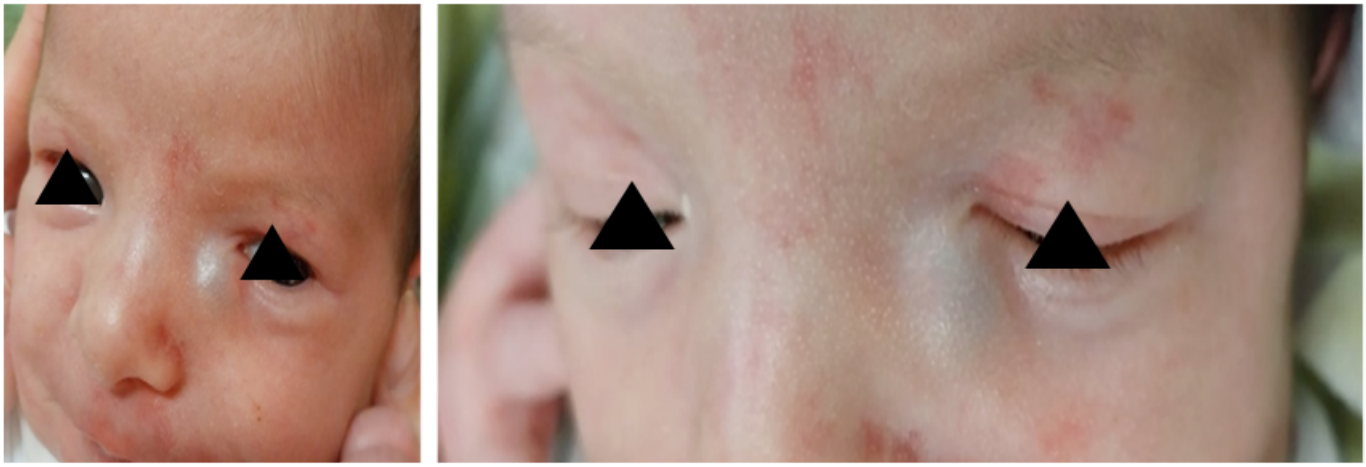

Figure 1: Pre-operative view of the 13 days old new-born.

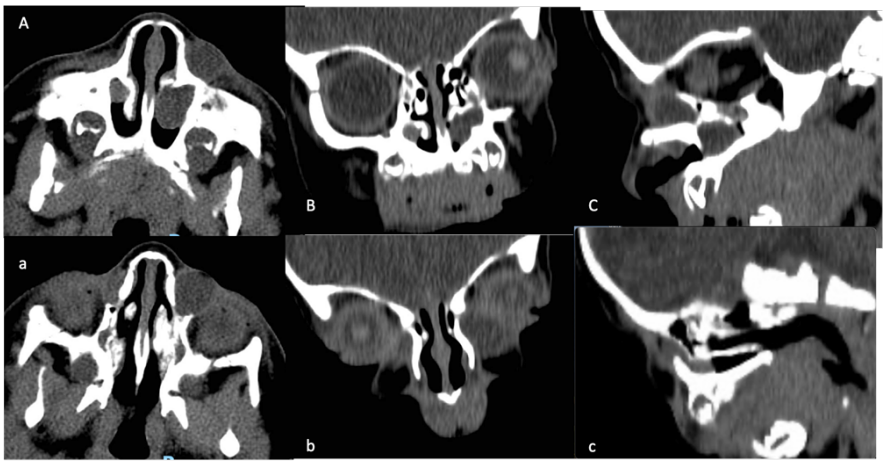

Figure 2: Axial (A, a), coronal (B,b) and sagittal (C,c) view CT scans showing the bilateral hourglass-shaped dacryocystocele.

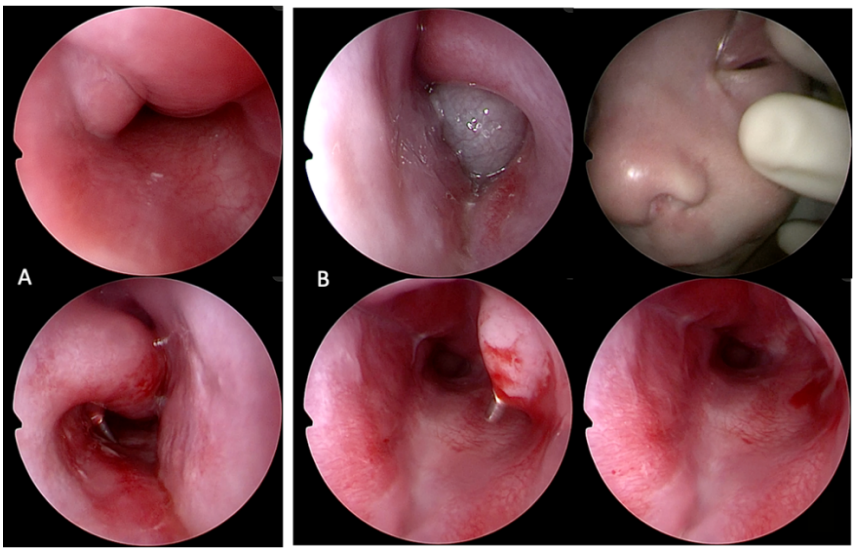

Figure 3: Endoscopic view of the nasal cavities during surgery: (A) right nasal fossa with the small nasal cyst drained and marsupialised with nasolacrimal duct probe; (B) left nasal fossa with near complete obstruction due to a mucocele in the inferior meatus displacing superiorly the inferior turbinate, incision and removal of the nasal cyst followed by inferior canaliculus probing and final view of the patent nasal fossa.

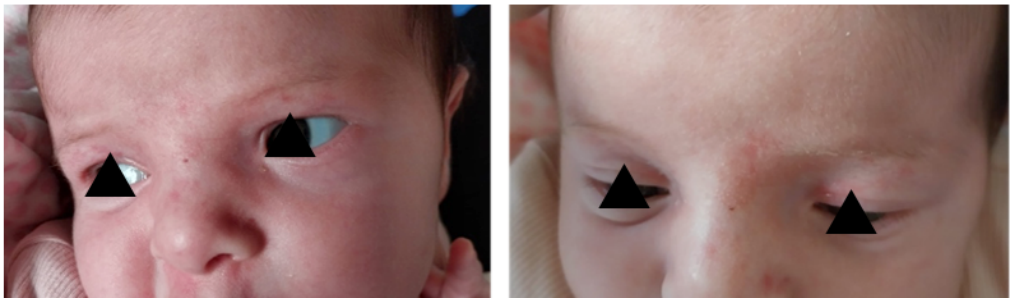

Figure 4: 10 days post-operative view of the patient.

Discussion

In new-borns, masses that might obstruct the nasal passages include hemangiomas, encephaloceles, nasal gliomas, neurofibromas, lymphangiomas, and nasolacrimal duct mucoceles [1]. Bilateral nasolacrimal-duct mucoceles (NLDMs) are extremely rare. However, bilateral mucoceles involving the lacrimal sac have been previously described [6]. NLDMs may be congenital or, less commonly, may develop secondarily to inflammations and oedema [1]. Other causes include abnormal folds in the mucosa and abnormal development of cartilage and bone of the nasolacrimal bone (3). During embryogenesis, the nasolacrimal duct arises from a linear thickening of the ectoderm in the naso-optic fissure [7,8]. In the third intrauterine month, canalisation of the ectodermal lining of the nasooptic fissures begins (4). In the proximal part, the canaliculi are highlighted, connecting the tiny orifices with the lacrimal sacs through Maier's sinuses. The distal portions will form the nasolacrimal ducts. During the sixth month of embryonic life, the canalisation begins, which is completed in the following months after birth [3]. Most authors maintain that maturation occurs segmentally from the ocular portion towards the nasal compartment. Therefore, the portion of the nasolacrimal duct would appear to be the last to be canalised [6,9,10]. The Hasner valve arises from a thin mucous membrane between the nasolacrimal duct's distal portion and the inferior meatus.

At birth, an imperforate Hasner valve is a common finding, with a frequency ranging from 6% to 73% [7,11,12]. At the time of birth, inspiratory effort and crying can cause rupture of the mucous membrane, therefore favouring the opening of the Hasner valve in a unidirectional manner [5]. Approximately 6-73% of new-borns have lacrimal duct stenosis that resolves spontaneously within the first year of life [1,5]. If the obstruction persists at any level of the nasolacrimal drainage apparatus, stagnation of secretion causes the appearance of lacrimal sac mucoceles, while a more distal blockage can cause NLDM. Suppose the little new-born has proximal and distal stenosis of the lacrimal duct at birth. In that case, it is called amniotocele, associated with enlargement of the lacrimal sac without signs of inflammation containing amniotic fluid [9,2,3]. On the contrary, if epithelial debris with a mucosal component is present, it is called a mucocele. Differential diagnosis does not change treatment. A mucocele of the nasolacrimal apparatus can extend superiorly and caudally; from a clinical point of view, distal or proximal endonasal swelling is evident. At clinical examination, the new-born may present a cystic mass of the medial can thus, a dilation of the lacrimal duct and an endonasal submucosal mass located under the inferior turbinate [1]. The skin region may be bluish [3]. Complications may include epiphora, dacrocystitis, periorbital cellulitis, sepsis or, rarely, dyspnoea [2]. Among diagnostic tests, nasal endoscopy plays a very important role because it is a minimally invasive and low-risk test, even if the new-born must be sedated. Exploration of the airways and in particular of the nasal passages allows the evaluation of any endoscopic bilateral lesions that may require a diagnostic biopsy [13]. Among the instrumental tests, CT significantly differentiates the triad of cystic dilatation of the lacrimal sac, dilatation of the nasolacrimal duct, and an intranasal cystic mass described by Rand et al. [1]. Furthermore, it is possible to carry out a differential diagnosis with other lesions such as meningocele, encephalocele, dermoid, dacrocystitis and some neoplastic processes such as hemangioma and lymphangioma. CT can be associated with magnetic resonance imaging to differentiate a mucocele of the nasolacrimal duct from some of these other conditions, as well as having the advantage of not exposing the child to ionising radiation with more excellent contrast resolution, especially in the suspicion of encephaloceles in new-borns. The MR scan was not performed because the combination of CT and clinical findings was considered diagnostic of a nasolacrimal duct mucocele. The high frequency ultrasound (US) can be very useful as it is a rapid, reliable, non-invasive examination and allows us to monitor the evolution of the pathology both in terms of size and vascularity for prevent serious infectious complications [14]. Moreover US can be useful to assess the cystic component of the dacryocele but do not provide useful information concerning eventual nasolacrimal drainage system bony or cartilagineous anomalies, and in such a small baby is also not easy to perform [15]. Conservative treatment consists of the instillation of local drops and rigorous hygiene of the eyelid, with massage of the lacrimal sac and only in case of infection the use of antibiotic eye drops. Conservative treatment is recommended for up to 12 months because from 14.2% to 96% of cases this pathology resolves spontaneously. If the problem persists within 12 months, surgical treatment should be considered [16]. Surgical treatment in new-borns is rare, but it is mandatory when complications such as dacryocystitis, cellulitis, large cyst causing astigmatism and narrowing of the lid fissure, respiratory difficulty caused by a nasal cyst and non-resolution of the cyst after a short trial period of massage. A nasal examination should be performed to exclude the coexistence of a nasal cyst responsible for respiratory distress, as the infants are nasal breathers [15,17,18]. The presence of nasal component requires an endoscopic approach that can be difficult due to the small dimensions of the nostrils and nasal fossa. Thanks to our experience in dealing with choanal atresia, we successfully treated the 13-day-old baby using a pediatric endoscope (2,7 mm diameter 0°) and suction and small instruments generally used for ear surgery. Bilateral nasolacrimal duct probing should be considered in unilateral cases because of the high incidence of occult contralateral involvement. Di Furia et al [19] marsupialized the lesion via endoscopic-assisted trans-nasal approach with the use of microdebrider.The nasolacrimal duct was cannulated to confirm patency. In our case, the external part was not evident, but the CT scan revealed the nasal mucocele

at the level of the inferior meatus.

Conclusion

Congenital dacryocystoceles are commonly associated with intranasal mucoceles, dacryocystitis, and pre-septal cellulitis. Respiratory distress is common in bilateral cases. Identifying these lesions is essential to plan immediate adequate therapy to prevent neonatal complications. Effective treatment requires timely recognition of the entity. The radiological evaluation with a CT scan plays a crucial role in diagnosing and differentiating this entity from other conditions, which may present as medial canthal masses. Surgical treatment aims to establish the patency of the nasolacrimal duct and usually leads to complete healing of the pathology.

Informed Consent: Informed consent was taken from the parents of the patient for publication of case report and pictures.

Conflict of Interest: The authors declare no conflicts of interest.

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