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Review Article

Neonatal airway obstruction in bilateral congenital dacryocystocoele: Case report and review of the literature

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ABSTRACT

Bilateral nasal obstruction due to simultaneous bilateral dacrocystocoeles is a rare and potentially life threatening condition. We present a five day old girl with this condition who presented with respiratory distress without any eye signs or symptoms associated with dacryocystocoeles. She was successfully managed with surgery and the clinical and radiological features of this condition are presented here with a review of the literature.

Contents

1. Introduction

Intranasal cysts secondary to bilateral congenital dacryocystocoeles are a rare and life threatening cause of upper airway obstruction in infants. While congenital obstruction of the nasolacrimal system is common, dacryocystocoeles are an uncommon and severe form of nasolacrimal duct obstruction [1]. This obstruction predominantly occurs at the valve of Hasner, the distal terminus of the system [2]. Most cases are unilateral and thus bilateral cases with intranasal extension are rare [3]. To our knowledge there are less than 25 reported cases in the literature. A dacryocystocoele expanding intranasally can result in respiratory distress as infants are obligate nasal breathers. This diagnosis should be kept in mind in cases of upper airway and nasal obstruction in neonates as prompt recognition is imperative. We describe a case of this condition successfully managed with surgery and a literature review of the diagnosis, management and prognosis.

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2. Case

A five day old girl presented to the Paediatric Emergency Department with increased work of breathing. She was born at term via elective lower section caesarian section due to a maternal fibroid. At birth she had some difficulty with breathing and was noted to be dusky. She was placed on an infant resuscitator but continually desaturated with attempted weaning so was transferred to the Neonatal Intensive Care Unit. Continuous Positive Airway Pressure (CPAP) was trialled empirically by the department as their standard of treatment for infants with respiratory distress before a diagnosis was established. Unfortunately there is no documentation available as to what type of mask was used. This was weaned successfully on the same day with no further respiratory distress and she was discharged.

Five days later she presented to hospital with 6–7 episodes per day of increased work of breathing, worsening with feeding. She had no apnoeic episodes, cyanotic spells, epiphora or rhinorrhoea.

On examination she was saturating at 95% on room air, and had no signs of respiratory distress. No misting was evident with a metal spatula. A 6 French feeding tube could not be negotiated through either nostril. In addition, eye examination was unremarkable specifically with no medial canthal swelling evident.

She was referred to our Paediatric Otolaryngology unit with an impression of choanal atresia. True complete choanal atresia was thought to be unlikely as she was not mouth breathing however a degree of choanal stenosis was suspected. A CT was performed revealing bilateral low density cysts present within the nasal cavity inferior to the inferior turbinate and patent choanae. These findings were consistent with bilateral dacryocystocoeles (see Fig. 1).

The patient was started on nasal decongestants and operative intervention was arranged. A combined Ophthalmology and Otolaryngology approach was utilised and she underwent an endoscopically assisted external drainage of bilateral dacryocystocoeles. Intravenous cephazolin was administered prophylactically. Both dacryocystocoeles were identified via nasendoscopy. Lacrimal probes were then used to dilate the lacrimal apparatus and advanced down to the floor of the nose where they were visible through the cysts endoscopically. Fluorescein was flushed through the duct delivering thick mucus from the now patent nasolacrimal ducts. Our team was on hand to perform cyst marsupialisation if needed but this was not required as further endoscopy revealed successfully decompressed cysts with patent nasolacrimal ducts represented by freely flowing fluorescein (see Fig. 2).

The patient had an uneventful course post-operatively and was discharged the following day on chloramphenical eye drops and oral Amoxiclav for 1 week. She was asymptomatic on follow up at one month and at six months. At this stage she has not required further nasendoscopy but has one further upcoming appointment.

3. Discussion

Our case report highlights the importance of early endoscopy and imaging in the diagnosis of this rare and potentially life threatening condition. A review of the literature reveals that defective nasolacrimal drainage occurs in 20% of newborns, indicating that congenital impatency of the system is common. However dacryocystocoeles, a severe form of nasolacrimal duct obstruction, occurs in only 2 in 1000 infants [1]. Approximately 90% are unilateral and thus bilateral intranasal cases are even less frequent [3]. This obstruction predominantly occurs at the valve of Hasner, the distal terminus of the system [2]. Multiple studies have demonstrated a female preponderance which is thought to be due to the nasolacrimal duct being narrower compared to males [3–6].

Congenital dacryocystocoeles typically manifest as a bluish, cystic mass in the medial canthal region. An intranasal mass in the anterior inferior meatus arising from the nasolacrimal duct is also often present [5]. Most patients have associated epiphora with mucopurulent discharge. Complications can include dacryocystitis and facial cellulitis [5]. A dacryocystocoele expanding intranasally can result in respiratory distress as infants are obligate nasal breathers. If there are bilateral intranasal dacryocystocoeles, this can potentially lead to life threatening airway obstruction [7]. To our knowledge less than 25 cases of bilateral intranasal extension that have been reported in the literature.

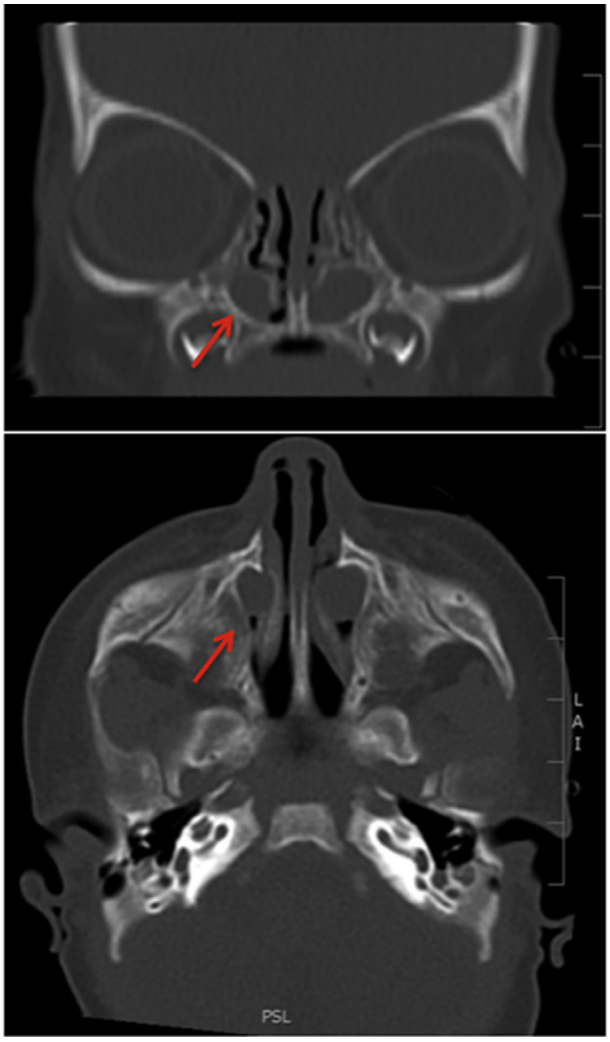

Fig. 1. Thin slice bony window CT sinuses. Coronal (top) and axial (bottom) views revealing bilateral intranasal low density cystic masses (red arrows) inferior to the inferior turbinate and in direct communication with enlarged nasolacrimal ducts bilaterally. Pyriform aperture and choanae patent with normal mid nasal diameter present. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

Examination includes both eye and nasal exams [5]. A thorough nasal examination is required to assess for a soft and compressible cystic mass arising from the undersurface of the inferior turbinate. Sometimes only redundant mucosa without obvious cyst formation is apparent [8]. Imaging can aid in diagnosis. For dacryocystocoeles, CT reveals a triad of lacrimal sac cystic dilatation, nasolacrimal duct dilatation and intranasal cystic mass [9]. CT can provide superior detail of the osseous nasal anatomy while MRI is useful if there is concern of an alternative diagnosis or intracranial communication [8].