Bilateral congenital dacryocystocele complicated with acute dacryocystitis

Dacriocistocele congênita bilateral complicada com dacriocistite aguda

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ABSTRACT

Congenital dacryocystocele is an uncommon entity that results from a malformation of the nasolacrimal system, occurring predominantly in neonatal females. It may resolve spontaneously but can be associated with potentially serious complications as acute dacryocystitis, requiring referral to an ophthalmologist. Bilateral cases of congenital dacryocystocele are rarely reported. We present a case of acute dacryocystitis occurring in a female newborn with bilateral congenital dacryocystocele who presented with bilateral epiphora and a mass in the right medial canthus since birth. Computed tomography revealed a bilateral soft tissue lesion in the medial canthus of the ocular globe. Dacryocystocele progressed to secondary infection on the right and patient developed acute dacryocystitis. She was admitted to the hospital for intravenous antibiotic therapy followed by the nasolacrimal system probing. This case report is also important to address the management of congenital dacryocystocele, and the decision to carry out the most suitable treatment, considering the diverse therapeutic options.

RESUMO

A dacriocistocele congênita é uma entidade incomum, que resulta de uma malformação do sistema nasolacrimal, ocorrendo predominantemente em recém-nascidos do sexo feminino. Pode se resolver espontaneamente, mas também pode estar associada a complicações potencialmente graves, como dacriocistite aguda, necessitando de encaminhamento a um oftalmologista. Raramente são relatados casos bilaterais de dacriocistocele congênita. Relatamos um caso de dacriocistite aguda acometendo um recém-nascido do sexo feminino. Ele apresentava dacriocistocele congênita bilateral, que apresentava epífora bilateral e uma massa no canto medial direito desde o nascimento. A tomografia computadorizada revelou lesão bilateral de partes moles no canto medial do globo. A dacriocistocele apresentou infecção secundária à direita, e a paciente desenvolveu dacriocistite aguda. Ela foi internada no hospital para antibioticoterapia intravenosa seguida de sondagem do sistema nasolacrimal. Este relato de caso também é importante para abordar o manejo da dacriocistocele congênita e a decisão de realizar o tratamento mais adequado, considerando as diversas opções terapêuticas.

INTRODUCTION

Congenital dacryocystocele is an uncommon anomaly affecting 0.02% of the newborns and 0.1% of the infants with nasolacrimal duct blockage. It is believed to occur because of concomitant upper obstruction of the Rosenmüller valve and lower obstruction of the Hasner valve of the lacrimal system, resulting in dilatation of the lacrimal sac and mucoid fluid accumulation within the nasolacrimal system. This condition is usually present at birth. It is usually unilateral, more frequent in Caucasians and females, and there is a familiar predisposition.[1,2]

The diagnosis is based on clinical features, which include a pink or blue mass in the medial canthal region of the eye, accompanied by epiphora since birth. However, when the diagnosis is uncertain, image tests such as computed tomography (CT), magnetic resonance imaging (MRI), and ultrasonography can be useful to show details of lacrimal system and the surrounding structures.[3]

Patients can experience spontaneous resolution of dacryocystoceles, but potentially serious complications such as acute dacryocystitis requires intravenous antibiotics or surgery to prevent life-threatening sepsis and meningitis.[4] We herein present a rare case of bilateral congenital dacryocystocele with complications caused by dacryocystitis.

CASE REPORT

A 20-day-old white female infant was brought to Pierre Ophthalmology Clinic with the history of a mass in the right medial canthal area accompanied by bilateral epiphora since birth. She was born healthy through Cesarean section at 38th week of gestation, and no other systemic diseases were presented. There was no history of respiratory distress. According to her mother, the patient presented with mild purulent discharge on the right eye, and mass gradually increased in size during the past week and had treated firstly with tobramycin eye drops prescribed by a primary care provider, without clinical improvement. Physical examination revealed a tender pink swelling under the right medial canthus (Figure 1), without purulent drainage by applying pressure over the lacrimal sacs. Anterior segment of both the eyes were normal. Computed tomography of the sinonasal region showed a bilateral soft tissue lesion medial to the globe, measuring 1.5cmx1.1cm on the right and 1.0cmx0.5cm on the left (Figure 2), with distention of the lacrimal sacs and dilatation of nasolacrimal ducts. No brain tissue herniation was seen in this region. The diagnosis of bilateral congenital dacryocystocele was made. The child received conservative treatment with Crigler massage, warm compress and oral azithromycin. However, 2 days later, she developed fever and acute dacryocystitis (Figure 3). That day, the infant was admitted to the hospital and treated with intravenous antibiotics (ceftriaxone and oxacillin) associated with gentle massage of the lacrimal sac. Patient improved with few days of therapy with a spontaneous rupture of the lacrimal abscess and leakage of purulent material on the right. There was resolution of the swelling after 7 days but epiphora still persisted. Endoscopy examination by otorhinolaryngologist was performed, and intranasal cysts were ruled out. Due the risk of new complications, surgical management was opted. She was successfully treated with nasal endoscopic-assisted probing of the lacrimal ducts under general anesthesia. The child was discharged after 48 hours from surgical treatment. Massage, warm compress, and eye drops continued to be used for 1 more week. The patient was checked one month later and was totally asymptomatic.

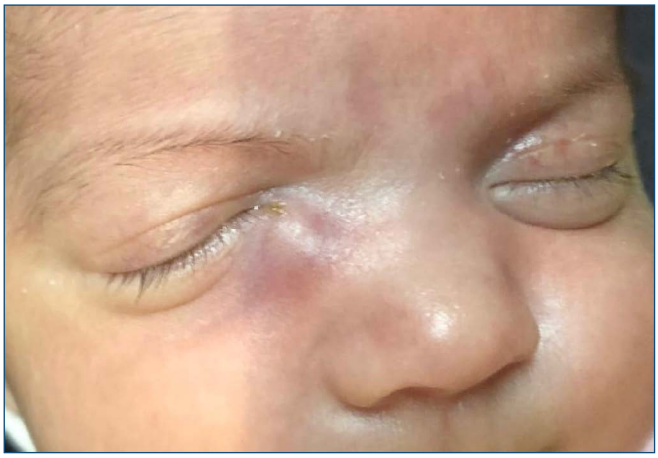

Figure 1. Patient with a pinkish swelling on the right side.

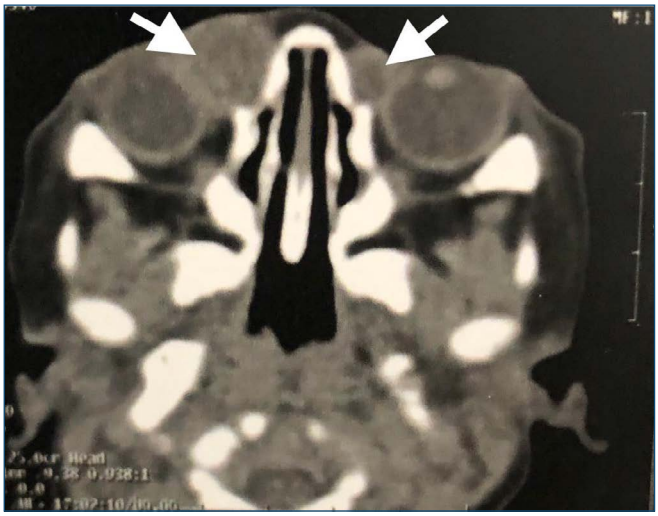

Figure 2. Axial computed tomography scan shows bilateral masses at the medial canthus region (arrows), with distension of the lacrimal sacs, confirming the presence of bilateral dacryocystocele.

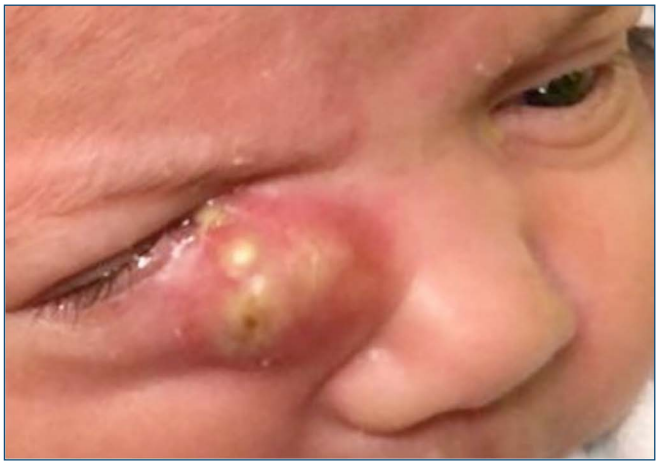

Figure 3. Newborn presenting with right acute dacryocystitis.

DISCUSSION

Dacryocystocele is mainly congenital and typically is presented in the first weeks of life. It results from the blockage of two sites in the nasolacrimal systema resulting in lacrimal sac enlargement, which may appear as a mass.[1-4] The diagnosis is made by clinical observation, but MRI, CT, ultrasound and nasal endoscopy are helpful to clarify and confirm the diagnosis. The absence of medial mass on examination does not exclude the diagnosis of bilateral congenital dacriocystocele.

Imaging is important to exclude other differential diagnosis of the affected region as encephalocele, meningoencephalocele, vascular malformations, dermoid cyst, lymphangioma, nasal glioma.[5] In our case, CT provided excellent resolution between bony structures and surrounding soft tissues and showed a cystic mass in the lacrimal sac and nasolacrimal duct enlargement bilaterally.

Bilateral dacryocystocele may extend intranasally causing airway obstruction or difficulty with deglutition. Lacrimal probing is recommended when it occurs.[6,7] Our case had a rare presentation of bilateral involvement in the absence of intranasal extension and no respiratory complications were observed.

Dacryocistitis is an acute infection of the nasolacrimal duct obstruction occurring in 20 to 75% of the patients with congenital dacryocystocele.[2,8] In neonates, it is an emergency condition, usually occurring within the first month of life and needs immediate treatment because of the high risk of septicemia and death, associated with the immature immune system of these patients.[9]

There is no consensus on the treatment of congenital dacryocystitis complicating dacryocystocele. Some authors[3,8,10,11] suggest initial treatment with antibiotics, while others prefer early surgical intervention,[4-7,12] especially in the presence of intranasal cyst. In a study of Davies et al.,[2] dacryocystitis resolved in 71% of patients without needing surgical intervention. In our case, despite the initial resolution of dacryocystitis by conservative measures and systemic antibiotics, there were bilateral dacryocystocele and epiphora. Early and late lacrimal probing has been shown to be safe and successful to treat nasolacrimal duct obstruction. The risk of recurrence is decreased when it is performed after antibiotic therapy is finished.[13,14] Our patient achieved a satisfactory resolution of the symptoms after surgery.

In conclusion, dacryocystitis is a possible complication from dacryocystocele that requires prompt treatment to prevent more serious problems. This case shows the importance of the cooperation of pediatrics, ophthalmologists, and otorhinolaryngologists for the correct diagnosis and medical management of congenital dacryocystocele and possible secondary complications. Nasal endoscopy may provide an important adjunct to the diagnosis and management of these infants.

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