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*Case Report*

# Congenital Bilateral Dacryocystocele

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## ABSTRACT

Congenital dacryocystocele is a rare entity and bilateral is rarer. It causes obstruction of lacrimal drainage system leading to swelling of the lacrimal sac. It may present at birth or soon after birth as a bluish swelling in the region of medial canthus. In bilateral cases there can be intranasal extension leading to respiratory distress. We present a rare case of bilateral congenital dacryocystocele with dyspnea due to intranasal extension. Imaging plays an important role in diagnosis. In our case computed tomography scan and magnetic resonance imaging revealed bilateral cystic swellings in the region of medial canthus adjacent to nasolacrimal duct causing its expansion with intranasal bulge. Dacryocystocele is managed conservatively and surgically. When a cyst appears in the region of lacrimal sac differential diagnosis of dacryocystocele should be considered.

***Keywords:*** dacryocystocele; lacrimal; congenital; canthus; bilateral.

***Key message:*** Bilateral congenital dacryocystocele is rare and can be identified along with its extent on imaging by CT (Computed tomography) and MRI (Magnetic resonance Imaging). On CT it appears as cystic medial canthal bulge, dilated nasolacrimal sac and nasal cavity bulge, On MRI it appears hypointense on T1-weighted images, hyperintense on T2-weighted and STIR sequence. Post-contrast studies may show minimal enhancement.

## INTRODUCTION

Congenital dacryocystocele is a rare occurrence due to nasolacrimal duct obstruction. Subsequently, there is accumulation of fluid leading to swelling over medial canthus. It is important to exclude intranasal extension leading to respiratory distress. CT (Computed tomography) and MRI (Magnetic resonance imaging) features are diagnostic.

## CASE HISTORY

10 month old female infant presented with history of painless, non tender swelling below the medial canthus bilaterally since birth. The swelling did not show any significant change in size since it was noticed. Infant had difficulty in breathing since few weeks after birth. There was no watering or discharge from either of the eyes. On examination, there were bilateral palpable fluctuant blue masses in the medial canthi. Anterior segments of both eyes were normal. Infant was dyspnoeic. There was no regurgitation of fluid on compression. Baby was born full term with a normal antenatal history. After clinical examination, radiological investigations were advised following which it was diagnosed as dacryocystocele. Imaging

findings on CT revealed two structures in anteromedial aspect (medial canthus) of both orbits in the region of nasolacrimal duct and lacrimal sac expanding it and appearing slightly hyperdense to the globe bulging into the nasal cavity (Figure1). On MRI, the bilateral anteromedial structures appeared hyperintense on T2-weighted and T1-weighted fat saturated images suggestive of cystic lesions. (Figure2). Infant was given oral antibiotics and topical antibiotic drops as a preventive measure with instructions of massage. On follow up after 1 week there was no resolution. Surgical management was opted. Under anaesthesia, following punctal dilatation, endoscopic nasal examination was performed. After locating the bilateral cystic masses, they were removed endoscopically and sent for histological examination. Post operative follow up showed resolution of both the medial canthal masses along with symptoms of dyspnoea. Histological examination confirmed the presence of benign lacrimal sac tissue. The patient was discharged with an advice of regular follow up for next 6 months. Currently, there is no evidence of recurrence.

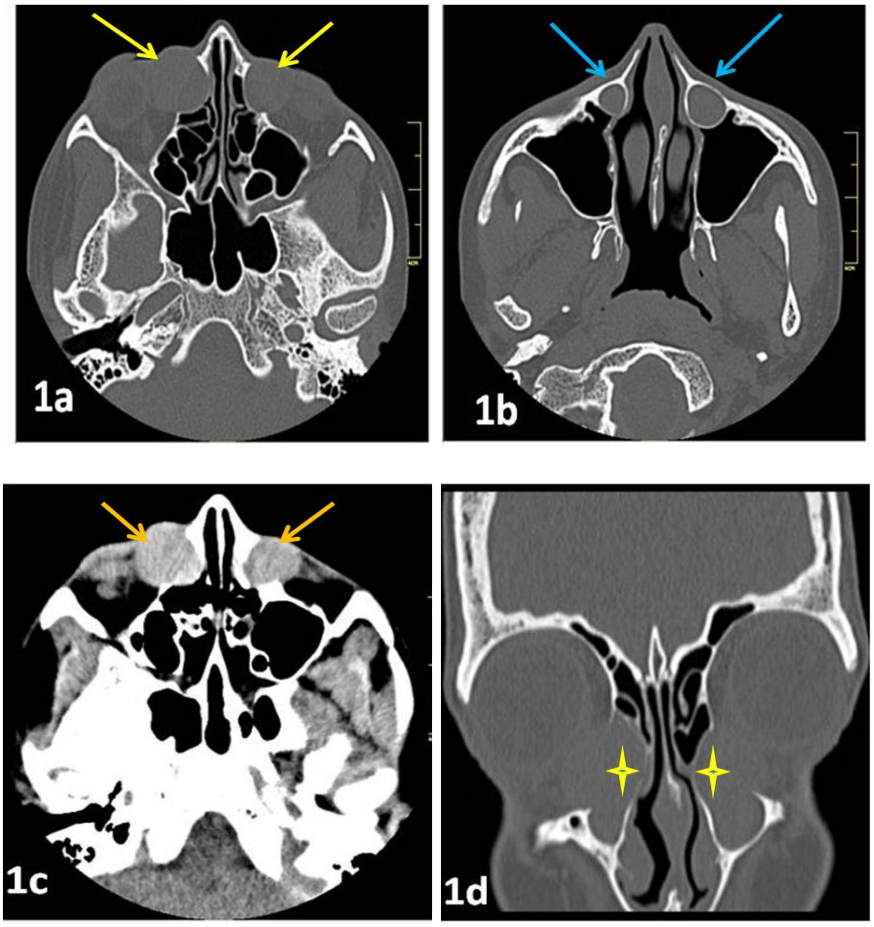


**Figure1: 10 months old female infant with bilateral dacryocystocele.**

**Findings:**

- **Figure 1a:** Axial CT images in bone window settings reveals two structures within medial canthal region of bilateral orbits adjacent to the nasolacrimal duct (yellow arrows) appearing almost isodense to muscles and slightly hyperdense to globe. The lesions are mildly bulging into the nasal cavity. The right side lesion is bigger than left. The masses measure 16x24x20mm on the right side and 14x18x15mm on the left side in superior-inferior, antero-posterior and transverse diameter respectively with 20-30 HU.

- **Figure 1b:** Axial CT images in bone window settings from a section inferior to the image 1a reveals two structures in the region of nasolacrimal duct (blue arrows) causing enlargement of nasolacrimal canal. The masses measure 16x24x20mm on the right side and 14x18x15mm on the left side in superior-inferior, antero-posterior and transverse diameter respectively with 20-30 HU.
- **Figure 1c:** Axial CT images in soft tissue window settings reveals two structures within anteromedial aspect of bilateral orbits adjacent to the nasolacrimal duct (orange arrows) appearing almost isodense to slightly hyperdense to muscles, likely due to high proteinaceous content.
- **Figure 1d:** Coronal CT images in bone window settings reveals two structures in the medial canthal region of bilateral orbits (yellow stars) causing enlargement of lacrimal sac fossa and nasolacrimal canal.

(Ct machine-16 slice Toshiba Aquilion ST-0.mm, mA-200, kVp- 120) .

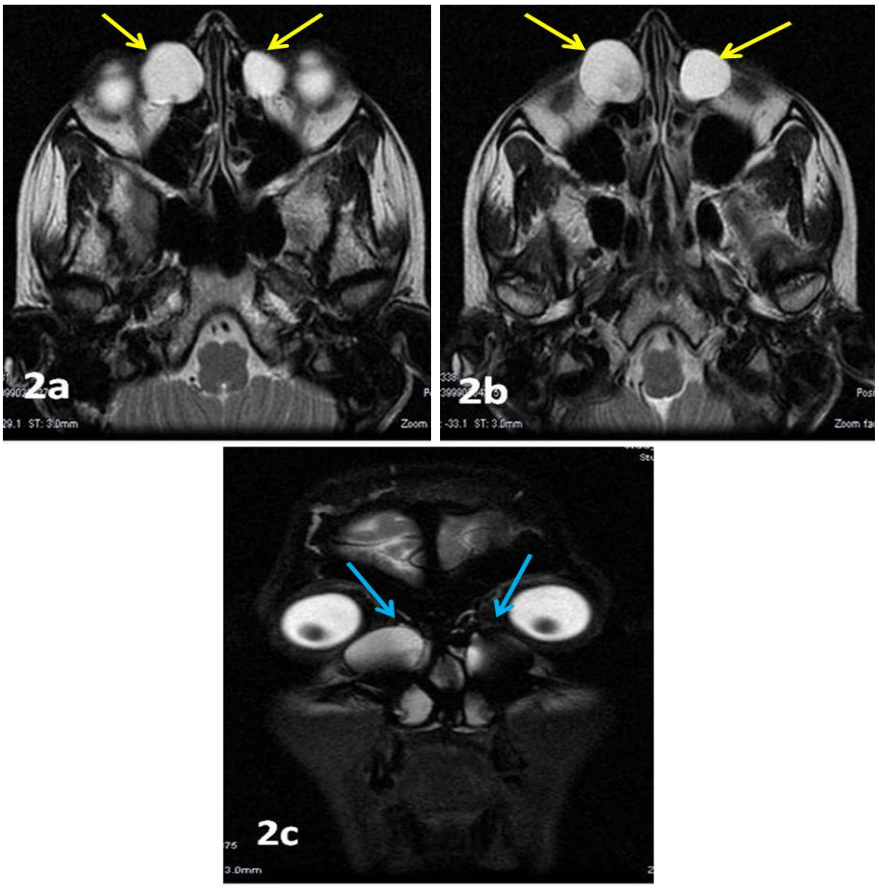


**Figure 2: 10 Months old female infant with bilateral dacryocystocele.**

**Findings:**

- **Figure 2a, b:** Axial T2 weighted images reveal homogeneous, hyperintense cystic masses in the anteromedial aspect of bilateral orbits in the region of lacrimal sac and nasolacrimal duct (yellow arrows). The masses measure 16x24x20mm on the right side and 14x18x15mm on the left side in superior-inferior, antero-posterior and transverse diameter respectively. In the inferior section there is extension of the lesion into bilateral nasolacrimal canals which are enlarged. 1.5T GE Signa Echospeed LX.TR- 4140, TE- 91.72, ST-3.mm.
- **Figure2c:** Coronal T2 weighted fat saturated images reveal two

homogeneous, hyperintense cystic masses in the anteromedial aspect of bilateral orbits in the region of lacrimal sac and nasolacrimal duct mildly bulging into nasal cavity (blue arrows). The masses measure 16x24x20mm on the right side and 14x18x15mm on the left side in superior-inferior, antero posterior and transverse diameter respectively. 1. 5T GE Signa Echospeed LX.TR- 5440, TE-86.223, ST-3.0mm.

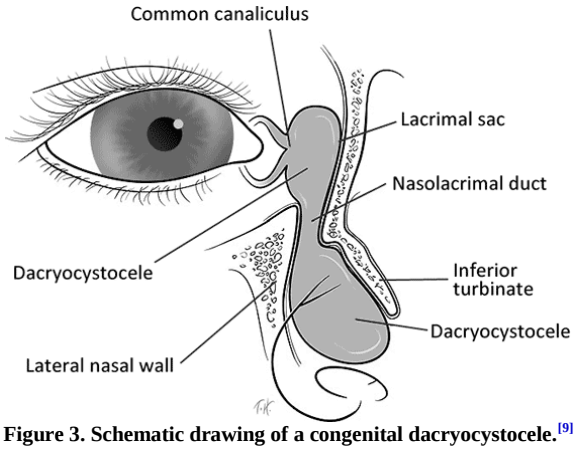


**Figure 3. Schematic drawing of a congenital dacryocystocele.**[9]

## DISCUSSION

Congenital dacryocystocele is a rare condition of nasolacrimal duct obstruction affecting new born and bilateral dacryocystocele is rarer. It occurs in 0.08% to 0.1% of newborns, 25% are bilateral. [1] It is more common in females and non-hispanic whites, while familial cases have been described only sporadically. [2]

It is a benign condition. Bilateral involvement may have intranasal extension which might be severe enough to cause obstruction resulting in neonatal respiratory distress requiring surgical intervention. [3] It appears as a swelling with bluish discolouration at birth or soon after, below and nasal to medial canthus. It is often misdiagnosed as a vascular lesion. [4] Congenital type is known to disappear in utero or spontaneously at birth. It is sometimes known to persist and would subsequently resolve during the neonatal period with conservative or surgical management. [5] It is commoner in females, could be due to narrower nasolacrimal duct than males. [6] Various causes of dacryocystocele include congenital deformities, trauma, primary and recurrent tumors affecting the nasolacrimal duct, idiopathic blockage of the nasolacrimal duct and iatrogenic causes including treatment of head and neck cancer in the sinonasal region. [7,8]

Dacryocystocele is caused due to blockage of the lacrimal duct in the nasal cavity at the level of inferior meatus. There is a valve of Hasner causing obstruction at this level which eventually undergoes perforation between 3rd to 6th months. Incomplete canalization of this valve leads to distal obstruction forming a dacryocystocele. [6] The obstruction of Rosenmuller valve, which normally prevents reflux from lacrimal sac to canaliculi, is another mechanism causing a dacryocystocele. [5] Thus, congenital dacryocystocele are due to obstruction proximally at the valve of Rosenmuller and distally at the valve of Hasner forming a one way valve allowing only fluid to enter in. Congenital type is typically known to occur in the third trimester as per the embryological changes. [10-12] There can be secondary infection due to stasis of the lacrimal fluid, location near the sinuses and the presence of lymphatic and vascular system. It could present as respiratory distress, dacryocystitis and facial cellulitis. [6] It can get infected most commonly by Staphylococcus aureus. [13]

It is important to look for any associated syndromes and structural abnormality. Sharony et al reported an association between dacryocystocele and genetic conditions such as Canavan disease and polycystic kidneys. [12] Polyhydramnios was associated in few cases. [20,14] Westbrook et al., reported recurrent bilateral dacryocystoceles in Wegener's granulomatosis. [15] It is important to

diagnose this condition due to the risk of intranasal extension causing obstruction to the nasal passages leading to neonatal respiratory distress requiring surgical intervention. [16] There are various opinions regarding treatment of dacryocystocele with either conservative or surgical treatment. Conservative treatment is with antibiotics, massage, nasal probing and also drainage if necessary. [6,17-19] However, if there is a delay in response, early surgical intervention must be applied or a quick first line surgical therapy especially with intranasal extension. [20,21] As per a study by Wong and Vander Veen, 28 of 42 patients presented with cellulitis or dacryocystitis and 36 of 46 affected eyes underwent surgical intervention. According to Schnall and Christian, 18% presented with dacryocystitis of 21 dacryocystoceles and 16 resolved with conservative therapy. 25% occurrence of dacryocystitis has been reported during the management of dacryocystocele. [6] As bilateral dacryocystoceles can cause obstruction of nasal passages leading to respiratory distress, surgical intervention is the treatment of choice. [17] The prognosis is good.

When the diagnosis of dacryocystocele is uncertain, diagnostic studies such as USG or CT or MRI imaging can be useful. [22] Ultrasound provides good information about the lacrimal sac, nasolacrimal duct and the surrounding structures. Antenatal 2D ultrasound is performed at 30weeks. It shows uni- or bilateral rounded cystic lesions inferiorly in the medial canthus region measuring few millimeters without vascularity or calcification with normal brain and facial structures. On 3D ultrasound precise location with a degree of intranasal extent and communication with orbits is clearly depicted. It also helps in better understanding for the parents. [6] However, other examinations such as dacryocystography, CT and MR imaging may be carried out to diagnose the condition and determine the extent. CT determines the bony involvement of the nasolacrimal canal and choanal atresia is easier to detect. [23,24] CT shows hypodense cystic medial canthal mass, dilated nasolacrimal sac, and submucosal nasal cavity mass. [25] MRI is the method of choice for soft tissue delineation and to study the relation with central nervous system before carrying out any interventions as it also differentiates solid tumor from this condition. MR imaging shows a mass with low intensity signal on T1-weighted images and high intensity signal on T2-weighted. [26] Post contrast studies may reveal minimal to no enhancement. It is essential to differentiate from other causes of duct obstruction or cystic dilatation such as ecephalocele, hemangioma and dermoid [1] [Use differential table]. On CT dacryocystocele appears as a cystic hypodense, fluid-filled structure with minimal enhancement. On MRI it appears hypointense on T1-weighted and hyperintense on T2-weighted sequences. On CT hemangioma appears as a well defined hyperdense mass with homogeneous contrast enhancement. [27] On MR, it appears hypointense on T1-weighted, hyperintense on T2-weighted and show diffuse homogeneous contrast enhancement. [27] On CT encephalocele appears as hypodense mass. On MR it appears hypointense on T1-weighted, hyperintense on T2-weighted and post contrast rim to homogeneous enhancement. [28] On CT dermoid appears hypodense, less dense than fluid. On MR it appears hyperintense on T1-weighted and hypointense on T2-weighted sequence. It shows post contrast rim enhancement. [29]

## CONCLUSION

Congenital dacryocystocele is an uncommon entity. Bilateral dacryocystocele is rarer. In such cases it is important to exclude intranasal extension leading to respiratory distress. Cases may be complicated or uncomplicated both requiring medical and surgical management. Examining paediatricians and neonatologists should be aware of its

presentation thus providing appropriate referrals for management. Radiologists should be familiar with the imaging findings.

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