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A Rare Case of Dacryocystocele Diagnosed by Antenatal Ultrasonography at 26 Weeks of Gestation

Parvathy Ganesan[1] Bijoy K. Balakrishnan[2] Meenu Batra Parasuram[2]

[1] Department of Fetomaternal Medicine, Pran Fertility and Well Women Centre, Thiruvananthapuram, Kerala, India
[2] Department of Fetomaternal Medicine, CIMAR Cochin Hospital, Kochi, Kerala, India

Address for correspondence Parvathy Ganesan, MS (OBGYN), Department of Fetomaternal Medicine, Pran Fertility and Well Women Centre, Thiruvananthapuram, Kerala 695011, India (e-mail: parvathyganesan86@gmail.com).

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Abstract

Dacryocystocele is a rare variant of obstruction of the nasolacrimal duct that results in a fluid-filled, closed sac. It often resolves by the spontaneous perforation of the distal membrane during the early neonatal period, resulting in drainage of the accumulated fluid. If persisting, this benign condition can be usually treated postnatally. If the cysts occur bilaterally, there can be an obstruction to the nasal passage due to their possible intranasal extension and might require surgical intervention postnatally to prevent or treat respiratory distress. Also, it may be a part of a few syndromes, which makes the early prenatal diagnosis very important. In this case report, we present a case of unilateral dacryocystocele reported as early as 26 weeks, 3 days of gestation detected by ultrasound that spontaneously resolved by 33 weeks. This is one of the earliest reported three-dimensional/four-dimensional ultrasound diagnosis of dacryocystocele.

Keywords
- dacryocyctocele
- prenatal diagnosis
- Inner canthus
- inferomedial
- complete resolution

Introduction

Dacryocystocele, also known as lacrimal duct mucocele or amniocele or amniotocele, is a rare variant of obstruction of the nasolacrimal duct that results in a fluid-filled, closed sac.[1] The lacrimal duct system develops in the second month of pregnancy, at around 6 weeks of gestation. It arises from the surface ectoderm that is located between the maxillary and the lateral nasal processes. It then begins to canalize at approximately 16 weeks of intrauterine life and is generally completed by 24th week of gestation. However, the canalization of the distal end of the drainage system at the level of the valve of Hasner may occur at birth or even few weeks later. Once the lacrimal punctum becomes patent, amniotic fluid gets filled in the distal end of the duct thereby causing distension of the duct system. There must be simultaneous distal anatomical obstruction at the level of valve of Hasner (isolated distal obstruction is called dacryostenosis) and a proximal fictional obstruction at the level of the Rosenmuller valve by the distended sac preventing its retrograde drainage through the puncta. This leads to the formation of dacryocystocele. It often resolves by the spontaneous perforation of the distal membrane during the early neonatal period, resulting in drainage of the accumulated fluid. Laterality and gender have not been reported significantly in prenatal studies. This benign condition can be usually treated postnatally. If the cysts occur bilaterally, there can be an obstruction to the nasal passage due to their possible intranasal extension. The respiratory distress developing, thereby often needs surgical intervention. Also, it may be a part of a few syndromes, which makes the early prenatal diagnosis very important. In this case report, we present a case of unilateral dacryocystocele reported as early as 26 weeks, 3 days of

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gestation detected by ultrasound that spontaneously resolved by 33 weeks. This is one of the earliest reported three-/four-dimensional ultrasound diagnosis of dacryocystocele.

Case Report

Mrs. S aged 22 years came for antenatal scan at our center. She was a third gravida with history of two first-trimester miscarriages for which she was evaluated earlier. The reports were all normal except for an abnormality in her karyotype (9qh+ variant). She visited us first at 15 weeks, 4 days of gestation, and underwent a scan that was normal. She did not opt for any genetic screening. Anomaly scan done at 22 weeks showed normal anatomy of the fetus including face and the central nervous system. There was an echogenic intracardiac focus in the left ventricle. She came for a reassessment scan after a month, at 26 weeks, 3 days. The ultrasound demonstrated a unilateral cystic lesion measuring 5.5 mm × 6.7 mm × 8.5 mm in the right inner canthus suggestive of dacryocystocele (►**Fig. 1**,**2**,**3**). The facial profile and the intraocular anatomy looked normal with the eyes

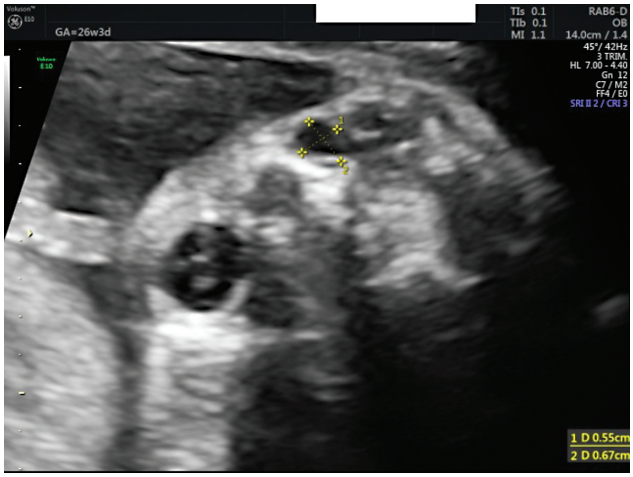

Fig. 1 Two-dimensional ultrasound demonstrating a cystic lesion in the right inner canthus suggestive of dacryocystocele.

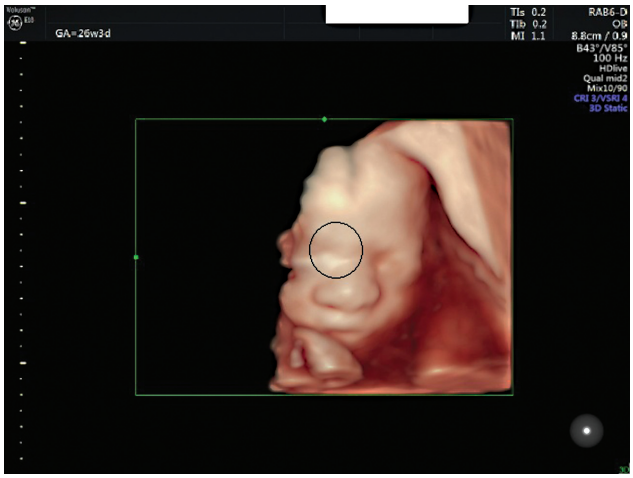

Fig. 2 Three-dimensional image showing dacryocystocele in the right inner canthus.

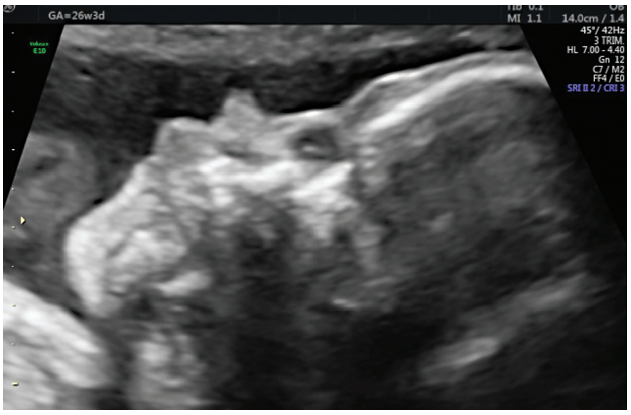

Fig. 3 Two-dimensional parasagittal image of fetal face demonstrating the dacryocystocele.

showing synchronous movement. The nervous system and brain anatomy were normal. She was counselled regarding the prognosis and the need for follow-up and the postnatal assessment. However, a follow-up scan at 34 weeks of gestation showed complete resolution of the lesion. She had premature rupture of membranes at 38 weeks gestation and delivered a live female baby weighing 2.8 kg. The baby had a normal APGAR (Appearance, Pulse, Grimace, Activity, Respiration) score and did not have any external deformities or swelling in the inner canthus.

Discussion

The prenatal diagnosis of congenital dacryocystocele has been already described. However, literature review shows only a few reports, with the earliest diagnosis made in the 27th week of gestation.[2] As mentioned before, the potential importance of the diagnosis is the possibility of intranasal extension thereby causing obstruction to the nasal passages (especially if there are bilateral cysts) and may cause neonatal respiratory distress for which surgical intervention is needed.[3] Neonates being obligate nasal breathers, there is the risk of acute respiratory distress in the early neonatal period and a pediatrician should be present for delivery.[4] The other possible differential diagnosis (►**Table 1**) of periorbital masses often includes cystic teratomas, hemangiomas, encephalocele, dermoid cysts, nasal glioma, and rhabdomyosarcoma.[2]

When the mass is unilateral, it is often difficult to differentiate between dacryocystocele and other peri orbital lesions.

Dacryocystocele is visualized as a hypoechoic mass lesion located inferomedial to the orbit. It can be seen in parasagittal or coronal plane including the medial angle of the orbits and the nose. On the contrary, a hemangioma is cutaneous in origin and is usually located in the head or neck, and can be solid or septated.[5] It is often differentiated from dacryocystocele by its peculiar Doppler patterns.[6] Dermoid cyst is situated superolateral to the orbit and has a complex appearance and hyperechogenicity on ultrasound, often with areas of calcification. Anterior cephalocele is a mid-line defect and is accompanied by a defect in the calvarium, and usually hydrocephalus. Lymphangioma, neurofibromatosis, and rhabdomyosarcoma are the other masses in the orbit, but these solid lesions are

Table 1 Differential diagnosis of periorbital swellings

Pathology	Dacryocystocele	Cystic teratoma	Anterior cephalocele	Hemangioma
Location	Inferomedial to the orbit	Superolateral to the globe	Midline defect with calvarial defect	Head/neck
Solid/cystic	Cystic	Complex	Solid/cystic	Solid/septated
Vascularity	Absent	Absent	Absent	Present

very rare. Another rare diagnostic dilemma is mucocele of the nasolacrimal duct system and is almost never diagnosed prenatally.[7] The typical appearance of the dacryocystocele generally allows the clear ultrasonographic diagnosis of this pathology. Particularly important features involved in differential diagnosis include the size, location, echogenicity, time of appearance, and the characteristics identified in Doppler flow.[8] A possible intra cranial connection is often ruled out with the help of magnetic resonance imaging (MRI).[9]

Prenatal diagnosis of dacryocystocele is relatively less reported in literature. Rand and Walsh reported two cases diagnosed at around 30th to 36th weeks of gestation.[10,11] The earliest reported diagnosis is by Sharony et al (27th gestational week) in their series describing six cases of dacryocystocele, often accompanying some pathologies or syndromes like Canavan disease, dysplastic kidney, pyelectasis, and maternal diabetes.[2] Recurrent bilateral dacryocystocele was observed by Westbrook et al, in cases of Wegener's granulomatosis.[12,13] Kim et al did a large retrospective analysis describing an overall incidence of prenatal dacryocystoceles to be 0.43%. The incidence was higher in the early third trimester and thereafter decreased. About 76% cases of prenatal dacryocystoceles resolved at birth, and the gestational age at delivery was a clinically significant predictor of its postnatal persistence.[14] In general, the lacrimal sac and the nasolacrimal duct visibility in the third trimester was reported to be around 45% with the peak gestational age of reporting being around 32 weeks. Congenital dacryocystocele usually represents those above the 95th percentile of the size of the lacrimal sac (i.e., $>$5 mm). The detection around 27 to 28 weeks actually represents the physiological sequences of the canalization (around 24 weeks of intrauterine life) or the opening of lacrimal punctum (range: 16–25/26 weeks).[9] Even though MRI reports suggest early detection (as early as 24 weeks), it is not a routine investigation during antenatal period. Also, considering the cost and inconvenience and difficulty in availability, the detection of dacryocystocele by MRI is of questionable value. The postnatal resolution was reported in approximately 70% of cases diagnosed by antenatal ultrasound.[8] The main thing to be noted here is the good prognosis and the timely advice that should be given to the patient. On follow-up ultrasound, our case had complete resolution and that itself is a key message to be delivered to the patients.

Implications for Clinical Practice

Correct diagnosis of dacryocystocele is very important and the earlier diagnosis and accurate differentiation from the other similar pathologies along with proper counseling and follow-up would help to manage this condition. If not resolving antenatally, postnatal follow-up is warranted and rarely some interventions might be needed. Early detection also would help to look for other features of associated syndromes, if any. Though few suggest there is no increase in incidence of genetic disease or syndromic association with the fetuses with dacryocystocele, it needs counseling of the parents to optimize perinatal outcome.

Conflict of Interest
None declared.

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