

Dacryocystocele on prenatal ultrasonography: diagnosis and postnatal outcomes

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Purpose: To report the incidence of dacryocystoceles detected by prenatal ultrasonography (US) and their postnatal outcomes and to determine the factors associated with the postnatal persistence of dacryocystoceles at birth.

Methods: We retrospectively reviewed the prenatal US database at our institution for the period between January 2012 and December 2013. The medical records of women who had fetuses diagnosed with dacryocystocel larger than 5 mm were reviewed for maternal age, gestational age (GA) at detection, size and side of the dacryocystoceles, delivery, and postnatal information, such as GA at delivery, delivery mode, and gender of the neonate.

Results: A total of 49 singletons were diagnosed with a dacryocystocele on prenatal US, yielding an overall incidence of 0.43%. The incidence of dacryocystoceles was the highest at the GA of 27 weeks and decreased toward term. Of the 49 fetuses including three of undetermined gender, 25 (54%) were female. The mean GA at first detection was 31.2 weeks. The dacryocystocele was unilateral in 29 cases, with a mean maximum diameter of 7 mm. Spontaneous resolution at birth was documented in 35 out of 46 neonates (76%), including six with prenatal resolution. Multivariate analysis demonstrated that GA at delivery was a significant predictor of the postnatal persistence of dacryocystoceles ($P=0.045$).

Conclusion: The overall incidence of prenatal dacryocystoceles was 0.43%; the incidence was higher in the early third trimester and decreased thereafter. Prenatal dacryocystoceles resolved in 76% of the patients at birth, and the GA at delivery was a significant predictor of postnatal persistence.

Keywords: Fetus; Ultrasonography; Congenital abnormalities; Lacrimal duct obstruction

Introduction

A congenital dacryocystocele (mucocele or amniotocele) is a rare variant of a relatively common congenital nasolacrimal duct obstruction, accounting for only 0.1% of infants with congenital nasolacrimal duct obstruction. It refers to cystic distention of the lacrimal sac as a consequence of concomitant proximal and distal obstruction of the nasolacrimal drainage system by a functional or

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mechanical cause. It typically presents as a bluish swelling inferomedial to the medial canthus in the neonate. Potential complications of congenital dacryocystoceles include acute dacryocystitis, orbital cellulitis, and respiratory distress when these dacryocystoceles extend intra-nasally and form a nasal cyst in the inferior meatus [1–5].

Prenatally detected dacryocystoceles are benign lesions and can resolve spontaneously during intrauterine life or the immediate perinatal period. However, accurate prenatal diagnosis of the dacryocystocele is important, because it enables postnatal follow-up. There have been a considerable number of reports on the prenatal diagnosis of congenital dacryocystoceles. However, a majority of prenatally reported cases are isolated case reports or review articles [6–12]. We present our experience in a large series of dacryocystoceles identified by prenatal ultrasonography (US). The aim of this study was to report the incidence of dacryocystoceles detected by prenatal US and their postnatal outcomes and to determine whether there are factors associated with postnatal persistence at birth.

Materials and Methods

Patients and Data Collection

We retrospectively reviewed the prenatal US database at our institution for the period between January 2012 and December 2013. The requirement of informed consent from patients was waived by the Institutional Review Board because of the

retrospective nature of this study. Cases of fetal dacryocystoceles were identified by reviewing the reading of the report of each fetal US. Dacryocystoceles were identified in 53 fetuses during the study period. Of these, two were twins and two had dacryocystoceles smaller than 5 mm. Brugger et al. [12] suggested that lacrimal sac diameters less than 5 mm should be considered normal, and lacrimal sacs with diameters greater than 5 mm should be considered prenatal dacryocystoceles. Therefore, 49 fetuses with a dacryocystocele having a maximum diameter of more than 5 mm in a singleton pregnancy were included in this study. For each case, maternal age, gestational age (GA) at first detection from the first date of the last menstrual period, and US features of the lesion, such as maximum diameter, side, and associated abnormalities, were recorded. The diagnosis of a dacryocystocele was made by the detection of an anechoic- or hypoechoic-cystic lesion on the inner lower medial aspect of the orbit (Fig. 1). Medical records of each case containing delivery and postnatal information such as GA at delivery, delivery mode, and gender of the neonate were also reviewed. Postnatal US of the area of the lacrimal sac of the neonates was performed in 41 cases diagnosed as dacryocystoceles by using prenatal US.

Prenatal US examinations were performed by 10 experienced sonographers, each with more than 2 years of experience, using Voluson E8, Voluson E6, Voluson 730 Pro, or Voluson 730 Expert (GE Healthcare, Milwaukee, WI, USA) and iU 22 or HDI 5000 (Philips Medical Systems, Bothell, WA, USA) machines equipped with 2–7-

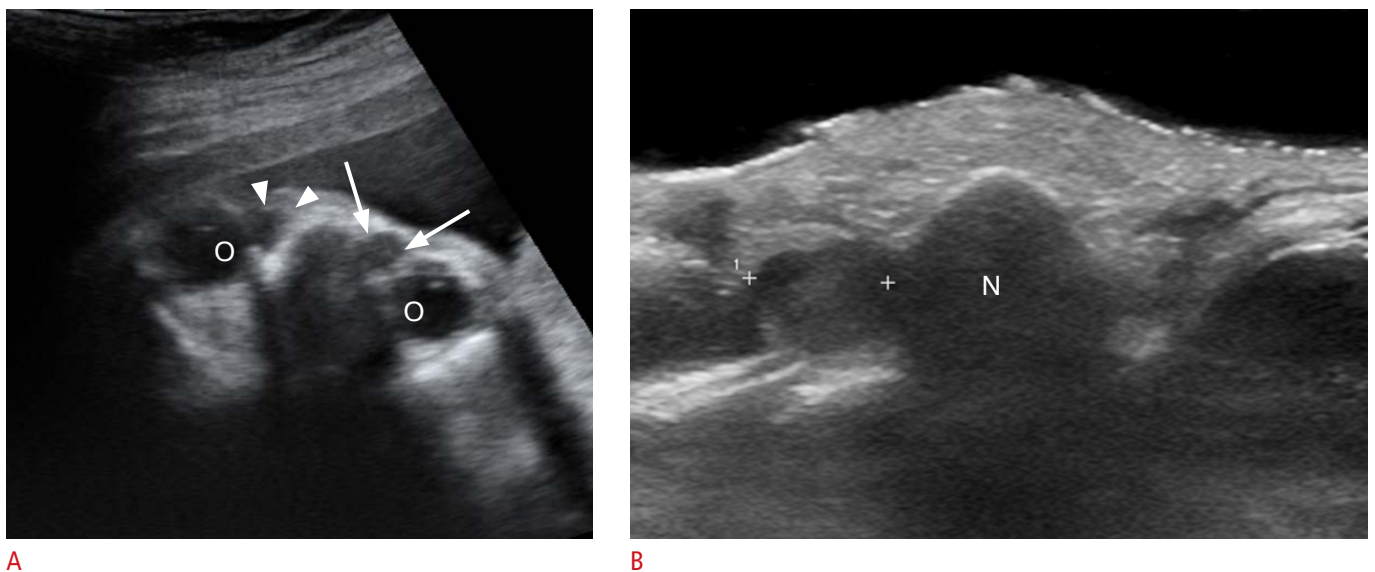


Fig. 1. Bilateral prenatal dacryocystoceles depicted on ultrasonography (US) with a persistent right dacryocystocele on postnatal US. **A.** Transverse view of the orbital area of a fetus at 32 weeks' and 1 day's gestation shows bilateral dacryocystoceles: a 7-mm hypoechoic lesion on the right (arrowheads) and another 8-mm hypoechoic lesion on the left (arrows) on the medial aspect of bilateral orbits. O, orbit. **B.** Transverse view of both orbital areas of the 1-day-old neonate on postnatal US shows a persistent hypoechoic lesion suggesting a dacryocystocele on the right (between the crosshairs), having a transverse diameter of 7 mm and no visible cystic lesion on the left. N, nose.

MHz curvilinear transducers. Postnatal US was performed by an experienced pediatric radiologist using a LOGIQ 9 scanner (GE Medical Systems) equipped with a 12-MHz linear transducer. The medial epicanthal areas were covered with a water-filled polyvinyl glove coated with normal saline.

Statistical Analyses

Because dacryocystoceles were diagnosed with prenatal US after 27 gestational weeks in this study, the incidence was calculated from the number of singleton pregnancies ($n=11,332$, total number of scans= $12,481$) that underwent prenatal US at or after 27 gestational weeks in the same period. As the number of pregnancies older than 36 weeks was small ($n=210$), they were grouped as 36+ gestational weeks for the statistical analysis. Eleven fetuses underwent routine follow-up US in the third trimester. In these cases, only the first study was used for the statistical analysis.

The data between the neonates with spontaneously resolved dacryocystoceles and the neonates with persistent dacryocystoceles on the postnatal US were compared using Pearson's chi-squared test or paired t-test. Variables with $P<0.05$ in the univariate logistic regression analysis were chosen for the subsequent stepwise multiple logistic regression analysis. A value of $P<0.05$ was considered statistically significant.

Results

Forty-nine singleton fetuses with dacryocystoceles having a maximum diameter of more than 5 mm were included in the study: 25 of these fetuses (54%) were female, 21 were male, and 3 were of undetermined gender at the time of US. Dacryocystoceles were identified at a mean GA of 31.2 weeks (range, 27.0 to 35.3 weeks). The dacryocystoceles were unilateral in 29 cases (59%) (right in 16 and left in 13) with a mean maximum diameter of 7 mm (range, 5 to 15 mm). Accompanying abnormalities were seen in two fetuses: one had suspected tetralogy of Fallot, and the other had preaxial polydactyly in the hand.

Overall, dacryocystoceles were detected in 0.43% of the third-trimester fetuses. The incidence of dacryocystoceles was 2.04% at 27 weeks and decreased thereafter to 0.19% at 35 weeks. The distribution of the study subjects and fetuses with dacryocystoceles according to GA is shown in Table 1. There was a peak at 32 weeks' gestation, which reflects our institution's practice of routine third-trimester screening. The prevalence of fetuses with dacryocystoceles for each age at the first diagnosis is shown in Fig. 2.

In 8 out of the 18 dacryocystoceles (44%) of the 11 fetuses who underwent prenatal follow-up US, the dacryocystocele (1 pair bilateral and 6 unilateral) disappeared at a mean age of 33.5 weeks

(range, 32.9 to 34.6 weeks). Of the remaining 10 dacryocystoceles, the size did not change in 5 at the follow-up US; it decreased in 4 by 1 mm on average and increased by 1 mm in 1.

Postnatal follow-up data were available in 46 of the 49 cases. Postnatal US for the lacrimal sac area after birth was performed in 41 neonates (median, 2 days; range, 0 to 14 days), while 5 neonates without any physical sign of dacryocystocele did not undergo postnatal US. Postnatal US showed persistent dacryocystoceles in 11 neonates (0.10% of the study pregnancies). In all, 35 out of 46 fetuses (76%) with prenatally diagnosed dacryocystoceles showed spontaneous resolution.

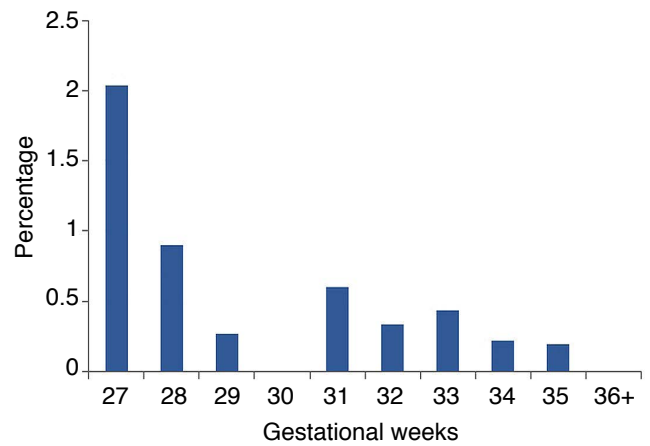
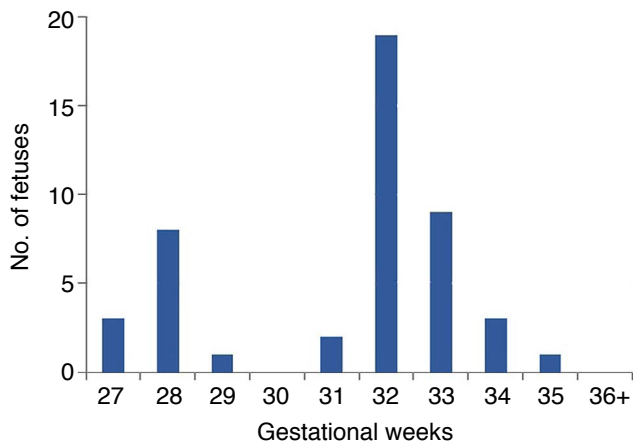
Of the 11 neonates with dacryocystoceles, 7 (64%) were girls and 8 (76%) had unilateral dacryocystoceles (right in 4 and left in 4). The mean maximum diameter of the dacryocystoceles was 11 mm (range, 8 to 15 mm). The mean GA of detection was 33.0 weeks (range, 32.1 to 35.4 weeks). Four babies were born by a cesarean section, and seven by vaginal delivery at a mean GA of 38.7 weeks (range, 36.9 to 41 weeks). One baby with bilateral dacryocystoceles showed chest retraction. No respiratory distress was recorded for the other 10 neonates. Among them, dacryocystoceles in eight babies resolved after massage and the application of topical antibiotics (7 within a month of age, and 1 after a month), two required nasolacrimal duct probing (one at postnatal 1 day, and the other at 15 days), and 1 was lost to follow-up. One baby was still receiving intermittent topical antibiotics at 16 months of age.

We compared the data between fetuses with spontaneously resolved dacryocystoceles and those with persistent dacryocystoceles on postnatal US among the postnatal data from the 46 patients

Table 1. Gestational age of the study population and prenatal dacryocystoceles

Gestational week	Total no. of US scans	No. of fetuses with dacryocystoceles
27	147	3 (2.04)
28	995	9 (0.90)
29	365	1 (0.27)
30	152	0
31	335	2 (0.60)
32	6,264	20 (0.33)
33	2,059	9 (0.43)
34	1,785	4 (0.22)
35	504	1 (0.19)
36+	210	0
Total no. of scans/ fetuses	12,481/11,332	49 (0.43)

Values are presented as number (%).
US, ultrasonography.



A

B

Fig. 2. Incidence of prenatal dacryocystoceles in 11,332 fetuses by gestational age.

Absolute number (y-axis on A) and percentage (y-axis on B) of the fetuses showing prenatal dacryocystoceles are demonstrated along with the gestational weeks (x-axis).

Table 2. Comparison of data between the resolution group and the persistent group on postnatal outcomes

Variable	Spontaneous resolution (n=35)	Persistent (n=11)	P-value
Maternal age (yr)	33.7	34.2	0.579
Gender of neonate			0.304
Male	17	4	
Female	18	7	
Delivery mode			0.739
Vaginal delivery	24	7	
Cesarean section	11	4	
GA at delivery (wk)	39.6	38.5	0.020
GA of first detection (wk)	31.2 (27.0–34.6)	33.0 (32.1–35.2)	0.001
Size (mm)	6.9	8.00	0.054
Laterality			0.103
Bilateral	17	3	
Unilateral	19	8	
Right	11	4	
Left	8	4	

Values are presented as number (range).

GA, gestational age.

(Table 2). There was a significant difference in the GA at first detection (mean GA at detection, 31.2 weeks vs. 33 weeks, respectively; $P=0.001$) and GA at delivery (mean GA at delivery, 39.6 weeks vs. 38.5 weeks, respectively; $P=0.020$) between the resolution group and the persistent group by univariate regression analyses. Interestingly, the size of the dacryocystoceles showed only

Table 3. Significance of variables predicting persistence of dacryocystoceles

Variable	Univariate analysis	Multivariate analysis	
	P-value	Adjusted odds ratio	P-value
Maternal age	0.579	1.088	0.580
Gestational age at delivery	0.020	0.402	0.045
Dacryocystocele size	0.054	1.353	0.322
Gestational age at detection	0.001	0.594	0.750

GA, gestational age; DCC, dacryocystocele.

a borderline-significant difference ($P=0.054$), with no significant difference in the laterality of the dacryocystocele, gender of neonate, or delivery mode. However, the multivariate analysis demonstrated that GA at delivery alone was significantly associated with the persistence of the dacryocystocele ($P=0.045$; adjusted odds ratio [OR], 0.402) (Table 3).

Discussion

The development of the lacrimal drainage system begins from the surface ectoderm located between the maxillary and the lateral nasal processes. Canalization of the lacrimal drainage system occurs simultaneously throughout the system, beginning at about 16 weeks of fetal life and is complete by 24 weeks. However, the distal end of the lacrimal drainage system at the level of the valve of Hasner may canalize at birth or even later. In this non-patent

distal end, the amniotic fluid fills and distends the nasolacrimal duct and the lacrimal sac after the patency of the lacrimal puncta. In the event of the early onset of mucus production by goblet cells, mucinous secretion may lead to distention of the lacrimal drainage system. Distention of the lacrimal duct and sac causes changes in the anatomic orientation of the proximal end of the lacrimal sac. This then acts as a check-valve system that only permits the fluid to enter into the sac, leading to further distention of the lacrimal sac. This cystic distention is designated as a dacryocystocele [7]. Fluid accumulation within the closed lacrimal drainage system enforces perforation of the membrane in the distal end of the nasolacrimal duct, the valve of Hasner, with resolution of the dacryocystocele.

The fluid-filled lacrimal sac and nasolacrimal duct were reported to become detectable after 24 weeks of gestation in a study using magnetic resonance imaging (MRI) [12]. The visibility of the lacrimal sac and the nasolacrimal duct in the third trimester was 45%, with a peak of 60% at 32 weeks, and decreasing thereafter to term, and lacrimal sacs larger than 5 mm, in the 95th percentile of the size range, are considered to represent congenital dacryocystoceles [12].

To the best of our knowledge, this is the largest study on prenatally detected dacryocystoceles. The overall incidence of dacryocystoceles on the prenatal US was 0.43%, which is in the range of prior reports. Sharony et al. [7] reported dacryocystoceles in 6 of 6000 (0.10%) prenatal sonographic surveys of low- and high-risk pregnancies. Using prenatal MRI, Yazici et al. [11] described 7 dacryocystoceles of 1,028 fetuses (0.68%), while Brugger et al. [12] showed 12 of 731 (2.76%). US is limited in revealing fetal structures overlaid by a bony structure. In cases of prone or oblique prone postures of the fetal head, the orbital area is difficult to evaluate due to shadowing by the facial bone or skull. MRI is advantageous at this point, and it has a better contrast resolution, and thus provides a more precise measurement and detection of the smaller dacryocystocele. This may explain the higher incidence in the evaluation with MRI. However, MRI was performed on fetuses nearly exclusively with US-detected abnormalities that might influence the formation of dacryocystoceles. Although MRI is increasingly utilized for fetal diagnosis, US is still the screening method of choice due to its superior spatial resolution, particularly in the second trimester, and its relatively low cost because it does not require manipulation of the patient and there is no potential for radiation exposure.

GA played a role in the level of dacryocystocele incidence in this study. The incidence was the highest (2.04%) at the GA of 27 weeks and decreased thereafter toward term. These results are in contrast with previous reports, which indicated that the dacryocystocele is more commonly detected after 30 weeks' gestation [8,9]; and the results may represent the intrauterine resolution of the dacryocystocele by the completion of the perforation of the

nasolacrimal duct. In fact, spontaneous intrauterine resolution of prenatal dacryocystoceles was noted in 44% of the eyes followed-up in our study. However, limitations in US visualization of the orbital area by posture or crowding of the fetal body at an advanced GA might be a factor for the decreased incidence in our retrospective study. Relatively less prominence of the dacryocystocele as compared to the enlargement of the fetal head as the GA advances may make detection more difficult. The fact that the number of scans before 32 gestational weeks was relatively small might be another factor responsible for the decreased incidence mentioned above. Therefore, further prospective investigations that take into consideration the visibility of the orbital area in an even distribution of the study population are needed.

Our finding that all of the fetal dacryocystoceles were diagnosed after 27 weeks of gestation is similar to previous reports [7,8,11]. All cases were detected after 27 weeks by US in studies with 6 and 10 cases [7,8], and after 26 weeks on MRI in a study with 7 cases [11]. The earliest lesion detection was reported at the GA of 25 weeks in a study using both US and MRI [13]. These results may reflect the timing of canalization of the nasolacrimal duct development, which is completed around 24 weeks of fetal life, or the timing of the opening of the lacrimal puncta, which ranges from 16 weeks to 25/26 weeks [12].

We showed that a prenatally detected dacryocystocele had a 76% chance of resolving spontaneously during the intrauterine period or at birth. The incidence of postnatal resolution was reported as 50% in a study with prenatal MRI and 70% in ultrasonographically diagnosed cases [8,11]. GA at delivery was the sole predictor of the persistence of dacryocystoceles in the multivariate analysis. With an adjusted OR of 0.402, the persistence of dacryocystoceles decreased by 40% for each increased week of GA from 36.9 weeks. This is correlated with the increasing frequencies of an open valve of Hasner after the seventh month, which reaches 66% in full-term fetuses [12] and may also be attributable to the decreased incidence of dacryocystoceles that we observed after 27 weeks. The finding that dacryocystoceles decrease in number as the GA increases is in agreement with the hypothesis that dacryocystocele formation occurs during the natural course of canalization in nasolacrimal duct development [4].

There was a significant difference in the GA of first detection between the resolution group and the persistent group in the univariate analysis. The earlier the first detection was, the more spontaneous was the resolution that occurred postnatally. Of note, all 17 of the dacryocystoceles diagnosed prenatally before 32 weeks spontaneously resolved at birth, although it was not statistically significant. We hypothesize that cases with earlier canalization may obtain earlier patency at the valve of Hasner by accumulating

more fluid that subsequently causes valve perforation [12]. There was no significant difference in the laterality of the lesion or the gender of the fetuses between the persistence group and the resolution group. Although a review of the literature showed a predominance of unilateral lesions in neonates [1,5,6,14], there was no predominance in the prenatal case reviews [6,8], which was also confirmed in our study. The preponderance of females with congenital dacryocystoceles reported in neonates [1,5,6] was not seen in the prenatal dacryocystocele cases that we examined. We also did not observe any difference in the mode of delivery; the assertion that mechanical decompression and the rupture of the valve of Hasner during vaginal delivery might be related to the resolution of the prenatal dacryocystocele [15] was not consistent with the findings of this study.

The birth prevalence of congenital dacryocystoceles in this study, 11 in 11,338 singleton pregnancies and about 1 in 1,030 births, is much higher than the previously reported value of 1 in 3,884 births during a 20-year study period [1]. This might be due to the fact that US examinations were performed on most neonates who had been diagnosed with dacryocystoceles prenatally in this study, whereas the previous report was a review of records of all patients younger than 5 years of age in a county. US may be able to delineate dacryocystoceles in mild cases that might be neglected by a physical exam alone. Another possibility is spontaneous perinatal resolution of dacryocystoceles in the natural course of nasolacrimal duct development because the patency of the distal end, where the valve of Hasner is located, may take up to a year to be fully developed in some. We performed US at a median age of 2 days postnatally; an earlier examination could result in the observation of a higher prevalence.

It has been reported that dacryocystoceles may be associated with other abnormalities or syndromes [7]. Reported accompanying malformation syndromes include ectrodactyly-ectrodermal dysplasia-clefting, Down syndrome, Canavan disease, and multicystic kidney disease. However, we found only two fetal abnormalities among the 51 prenatally diagnosed dacryocystoceles in this study: a suspected tetralogy of Fallot with follow-up loss and preaxial polydactyly in the hand, which was confirmed postnatally.

In conclusion, the overall incidence of prenatal dacryocystoceles was 0.43% on US; the incidence was the highest in the early third trimester and decreased thereafter. Prenatal dacryocystoceles resolved in 76% of the infants at birth, and the GA at delivery was a significant predictor of postnatal persistence.

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Conflict of Interest

No potential conflict of interest relevant to this article was reported.

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