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ULTRASOUND IMAGE OF THE MONTH

"Double eyes" sign of congenital bilateral dacryocystoceles

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Abstract Dacryocystocele is caused by nasolacrimal duct obstruction and results in cystic dilatation of the proximal part of the nasolacrimal duct, which is located inferomedial to the orbit, leading to fluid accumulation. It is important to consider that persistent congenital bilateral dacryocystoceles may cause neonatal nasal obstruction resulting in respiratory difficulty, and large dacryocystoceles may require surgical drainage. Ultrasonography demonstrates that congenital bilateral dacryocystoceles and normal eyeballs prenatally resemble two pairs of cystic "lesions" of different sizes. We herein present a case of prenatally diagnosed isolated congenital bilateral dacryocystoceles and propose the new name of "double eyes" sign for this rare condition to create an impact on medical students and residents.

Keywords Dacryocystocele · Lacrimal duct cyst · Prenatal diagnosis · Fetal ultrasound

Introduction

Dacryocystocele is caused by nasolacrimal duct obstruction, resulting in cystic dilatation of the proximal part of the nasolacrimal duct, which is located inferomedial to the orbit, leading to fluid accumulation [1, 2]. Well-circumscribed orbital anechoic or hypoechoic cysts of variable sizes that do not displace the eyeballs are typically observed [1]. An isolated dacryocystocele is not associated with aneuploidy, and most cases resolve without surgical intervention [2]. It is necessary to know that persistent congenital bilateral dacryocystoceles may cause neonatal nasal obstruction resulting in respiratory difficulty at birth, and large dacryocystoceles may require surgical drainage [1, 2].

Congenital dacryocystoceles resemble "extra eye" [2], and bilateral cases particularly resemble two pairs of cystic "lesions" of different sizes. Congenital bilateral dacryocystoceles constitute 25 % of all dacryocystoceles [2] and should be screened for the possibility of intranasal extension. "Double bubble" for duodenal atresia and "double bladder" for fetal intra-abdominal umbilical vein varix, for example, create an impact on medical students and residents to indicate fetal disorders [3, 4]. We herein present a case of prenatally diagnosed isolated congenital bilateral dacryocystoceles and propose the new name of "double eyes" sign, referring to the ultrasonographic feature.

Case report

A 32-year-old woman, $G_1P_1A_0$, consulted the Bern Forest Clinic complaining of amenorrhea at 6^{+2} weeks' gestation in a naturally conceived normal pregnancy. At the age of 29 years, she had delivered a male newborn weighing 2,710 g (appropriate-for-date, AFD) at 37^{+6} weeks' gestation by Cesarean section because of abnormal fetal presentation. Her genetic, family, and other previous histories were unremarkable. No abnormality was detected on routine obstetrical ultrasound as of 25^{+4} weeks' gestation.

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At 27^{+6} weeks' gestation, bilateral, uniloculated, anechoic and avascular cystic "lesions" were observed inferomedial to the eyeballs. At 30^{+1} weeks' gestation, both masses slightly enlarged and resembled two pairs of eyes: external pair of "large eyes" and internal pair of "small eyes," i.e., "double eyes" sign (Figs. 1, 2). No other morphological abnormality was observed. We followed up the size of the internal masses in sequential examinations. Both masses completely disappeared at 31^{+6} weeks' gestation and did not reappear thereafter. Thus, we diagnosed them as isolated temporary congenital bilateral dacryocystoceles that resolved spontaneously in utero.

At 37^{+5} weeks' gestation, an elective Cesarean section was performed because of the previous Cesarean section. This time, she delivered a female AFD newborn weighing 2,604 g with Apgar scores of 9 and 10 at 1 and 5 min, respectively. No morphological abnormality was observed around the bilateral neonatal medial canthi.

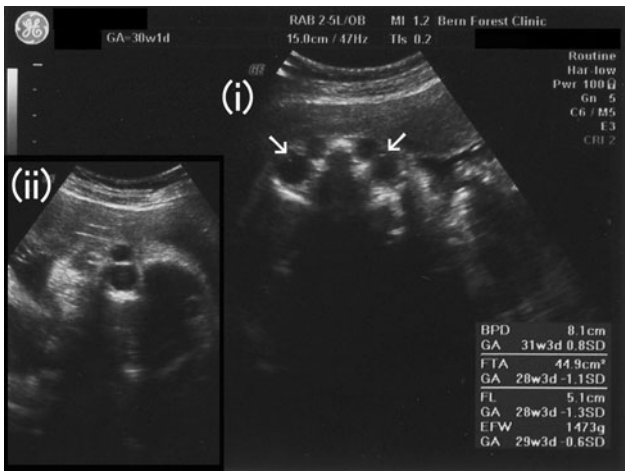

Fig. 1 Ultrasonographic findings at 30^{+1} weeks' gestation. *i* Axial view of the fetal head shows bilateral, uniloculated, anechoic and avascular orbital cystic "lesions" with thin walls inferomedial to the eyeballs. Lenses are observed in the eyeballs (*white arrows*), but not in the cysts. The right cyst is 7.4 mm × 7.8 mm and the left cyst is 7.6 mm × 8.1 mm in size. *ii* Sagittal view of the fetal head shows that there is no mass effect on the orbit

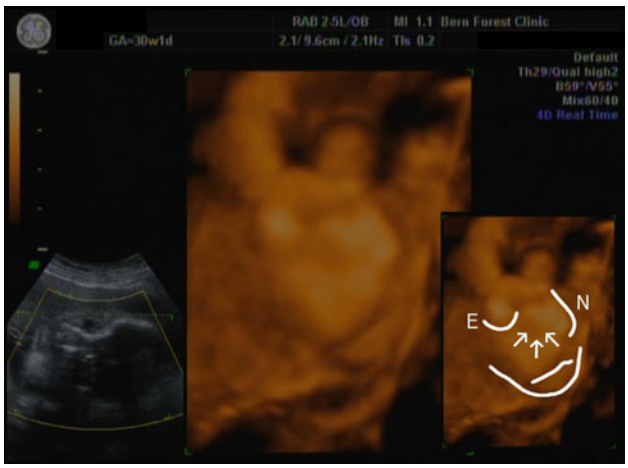

Fig. 2 Ultrasonographic findings at 30^{+1} weeks' gestation. Surface-rendered view of the fetal face shows a focal swelling mass between the upper nose and right eye (*white arrows*). *E* fetal right eye, *N* fetal nose

Discussion

"Double eyes" sign refers to congenital bilateral dacryocystoceles, representing two pairs of cystic "lesions" of different sizes; external lesions with lenses are eyeballs and internal lesions are dacryocystoceles. "Double" creates an impact on medical students and residents, as reported previously by authors in specialized fields; however, it does not represent the ultrasonographic finding in the case of a unilateral dacryocystocele. Proposing a new name for this rare disorder is important to place emphasis on the possible adverse neonatal outcomes of bilateral dacryocystoceles [5]. Because newborns are nasal breathers [2], pregnant women with fetuses complicated by persistent congenital bilateral dacryocystoceles should deliver in perinatal medical centers.

Dacryocystoceles are caused by obstruction of the lacrimal gland valves, i.e., Rosenmüller or Hasner valves, causing accumulation of mucus or amniotic fluid in the obstructed duct [2]. After birth, gray-blue-colored mucoid material in cystic swellings below the medial canthus is typically observed [2]. Congenital dacryocystoceles generally present after 30 weeks' gestation, with an incidence of 1 in approximately 4,000 live births [6]. Dacryocystoceles are benign self-limited entities, and approximately 50 % of cases resolve in utero and 85 % resolve in the first year of life [1, 2]. The present case showed typical characteristics of slow growth and spontaneous resolution in utero.

It is necessary to exclude the differential diagnoses such as amniotic band syndrome, facial hemangioma or lymphangioma, frontal or nasal encephaloceles, nasopharyngeal teratoma, proboscis, and retinoblastoma [1, 2]. For accurate diagnosis, connection to the amniotic membranes, avascularity, echogenicity, location, unilocularity, and spontaneous disappearance of the cyst are useful findings. Furthermore, three- or four-dimensional ultrasonography and fetal magnetic resonance imaging may provide better diagnostic findings; the former can provide better distinction from other fetal facial lesions, and the latter can delineate dacryocystoceles in a clear, detailed fashion [7, 8].

Conflict of interest None.

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