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Research Letter

Prenatal progressive cardiomegaly and functional pulmonary atresia on one fetus of monochorionic diamniotic twin pregnancy regardless of spontaneous resolution of isolated polyhydramnios

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Dear Editor,

A 32-year-old multiparous woman presented to our hospital for perinatal management of monochorionic diamniotic (MD) twin pregnancy at 26 weeks of gestation. The first transabdominal ultrasonography demonstrated that maximum vertical pocket (MVP) was 10.7 cm (twin A)/2.3 cm (twin B) and cardiothoracic area ratio (CTAR) was 37.4%/31.7%. Obvious tricuspid regurgitation (TR) and enlarged bladder were detected in twin A. No major structural anomaly and abnormal Doppler waveforms of the umbilical artery were detected in both fetuses. Because this case didn't meet the criteria of TTTS during the observation, fetoscopic laser surgery was not performed. Regardless of spontaneous normalized polyhydramnios at 28 weeks of gestation, severe TR was prolonged and CTAR continued increasing in twin A (Fig. 1). At 35 weeks of gestation, fetal echocardiography showed retrograde blood flow in the ductus arteriosus and extremely decreased forward blood flow through the pulmonary valve, which suggested functional pulmonary atresia (fPA). Repeat cesarean section was conducted at 37 weeks of gestation. Both were male and birth weight of twin A and B were 2902 g/2591 g, respectively. Echocardiography of twin A at birth demonstrated severe TR and slight forward blood flow through the pulmonary valve. Desaturation occurred in about 3 h after birth and prostaglandin E1 (PGE1) was infused, based on the diagnosis of fPA. PGE1 infusion was suspended on postnatal day 2. After that, he was alive without major complications other than moderate TR at the age of nine months.

Isolated polyhydramnios (I-Poly) was first reported by Chon et al. [1]; they reported an MVP with one twin sac $\geq$ 8 cm and the other sac > 2 cm and <8 cm in an MD twin pregnancy. Perinatal outcome of I-Poly with spontaneous normalized polyhydramnios is favorable because the intact survival of both twins at one month of age was reported as about 94% [2]. Right ventricular outflow tract abnormality (RVOTA) is reported as a cardiac abnormality occurring in MD twin pregnancy, especially complicated in the recipient (RT) of severe twin–twin transfusion syndrome (TTTS) [3,4]. Whereas, the characteristic of RVOTA of MD twin pregnancy with I-Poly has not been adequately known. Structural heart change of severe TR is well understood through many researches of adult's hearts [5]. The pathogenesis of the phenomenon of this case; cardiomegaly due to severe TR was progressive despite the resolution of the blood flow imbalance between the twins, was possibly that the tricuspid annular dilatation was caused by right ventricular or atrial expansion due to volume overload in a certain period of time; which caused increment of TR and the right ventricular or atrial expansion was escalated, falling into a vicious circle.

In conclusion, prenatal fPA with progressive cardiomegaly can occur even in MD twin pregnancy complicated by I-Poly regardless of spontaneous resolution of polyhydramnios. Even if the clinical condition such as amniotic fluid discordance is judged to have

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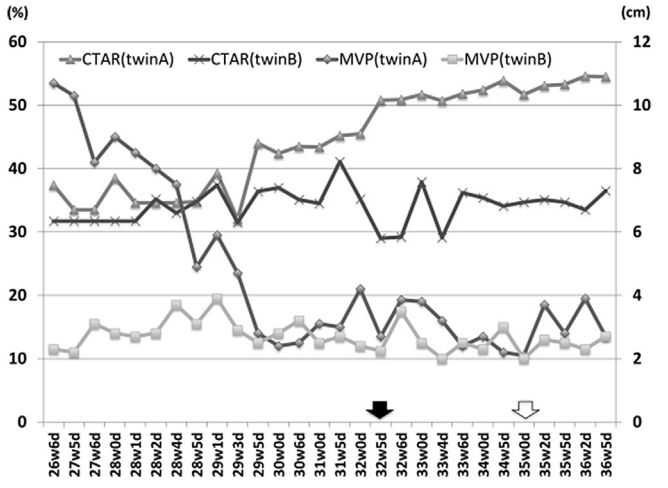

Fig. 1. Line graphs about the transition of both cardiothoracic area ratio (CTAR) (%) and maximum vertical pocket (MVP) (cm) of amniotic fluid volume respectively by transabdominal ultrasound. Black arrow shows the timing of emerging pericardial effusion. White arrow shows the timing of emerging retrograde blood flow of ductus arteriosus.

spontaneously improved, careful fetal cardiac evaluation and perinatal management should be performed bearing in mind immediate intervention for the neonate just after birth.

Conflicts of interest

The authors indicated no potential conflicts of interest.

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