



Research Letter

Three-dimensional HDlive image of *ectopia cordis* in a twin fetus at 9 gestational weeksYi Chang^a, Ming-Jie Yang^{a, c}, Peng-Hui Wang^{a, c}, Chih-Yao Chen^{b, c, *}^a Department of Obstetrics and Gynecology, Taipei Veterans General Hospital, Taipei, Taiwan, ROC^b Dianthus MFM Center Minquan, Dianthus Medical Group, Taiwan, ROC^c National Yang-Ming University, School of Medicine, Taipei, Taiwan, ROC

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Ectopia cordis is a congenital malformation in which the heart is abnormally located partially or totally outside of the thorax. The occurrence rate is eight per 1,000,000 births. Multiple gestations are associated with a greater risk of congenital anomalies than singleton pregnancies. Previously, the diagnosis has been reported as early as the 12th week of gestation [1]. Three-dimensional (3D) ultrasound has been useful in better defining the pathology of *ectopia cordis* [2]. We report a case of a twin fetus with *ectopia cordis* at 9 weeks' gestation. The woman was impregnated via *in vitro* fertilization. We have documented the prognosis of both fetuses and the mother.

A 36-year-old Taiwanese woman was initially presented at our reproductive medical center due to a barren marriage for a span of years. A series of fertility tests were executed. Male factor non-obstructive azoospermia was the diagnosis. Thus, *in vitro* fertilization with frozen embryo transfer was performed. Dichorionic diamniotic twin gestation with both fetal heart beats was detected for the first time at 5 weeks' gestation.

At 9 weeks' gestation age, fetal color Doppler ultrasound showed a bulging mass outside the thorax with blood flows of Twin B that strongly indicated *ectopia cordis* (Figure 1). 3D reconstructed images are shown in Figure 2. Ultrasound showed no specific structure abnormality of Twin A. We informed the woman of the above sonographic finding. After extensive counseling, the patient decided to carry on the pregnancy without fetal reduction. At 20 weeks' gestational age, ultrasound was performed. The ultrasound finding of Twin B included *ectopia cordis*, cleft palate, and cleft lip. The ultrasound of Twin A revealed no remarkable abnormality.

At 30 weeks' gestational age, water break with labor pain was noticed. The patient had a poor response to tocolytic agents and received a cesarean section 3 days after admission. Twin A, a female baby, was delivered with Apgar score 6 at 1 minute and Apgar score 8 at 5 minutes. The birth weight of Twin B was 1660 g, and the Apgar score of Twin B was 3 at 1 minute and Apgar score 2 at 5 minutes. Physical examination showed similar findings to the prenatal ultrasound exam: *ectopia cordis* and cleft palate and lip (Figure 3). Due to the fact that a do-not-resuscitate informed consent had been signed by the parents, Fetus B expired shortly after birth. Unfortunately, postpartum hemorrhage was noticed shortly after the cesarean section. We performed a subtotal hysterectomy to control the postpartum hemorrhage. She was then discharged under a relatively stable condition.

First-trimester prenatal diagnosis of fetal *ectopia cordis* in twin gestations using ultrasound was documented for the first time by Kristen et al [3] in 2009. In that case, sonographic diagnosis of combined *ectopia cordis* and holoprosencephaly was made at 12 weeks' gestational age. It might be easier to detect these with an

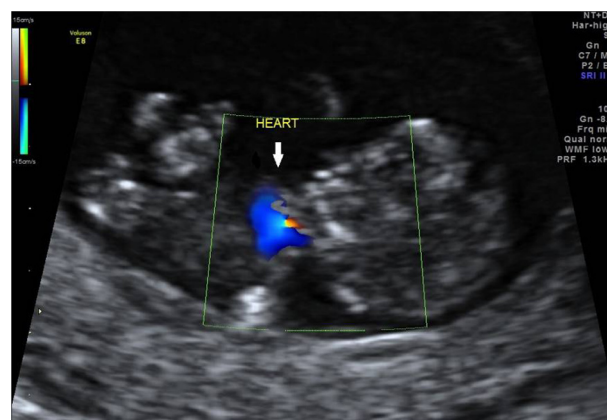


Figure 1. Fetal heart exposed outside the thoracic cavity of Twin B at 9 weeks' gestation. The anatomical structure appeared normal. Color Doppler scan confirmed the cardiac structure of the bulging mass.

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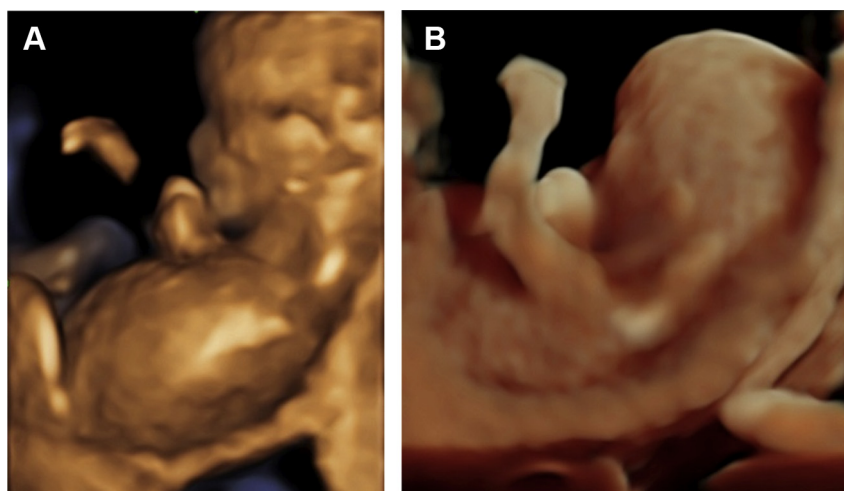


Figure 2. Three-dimensionally reconstructed image of Twin B. (A) Conventional three-dimensional reconstructed image and (B) three-dimensional high definition live image.



Figure 3. Gross picture of Twin B. *Ectopia cordis* and a cleft palate and lip are noted.

ultrasound because of multiple fetal anomalies. In 2013, Grigore [4] also described a case where a woman had a twin pregnancy and had one fetus with *ectopia cordis* diagnosed at 12 weeks' gestational age. Nevertheless, Grigore's [4] case did not show a postpartum gross picture due to selective fetal reduction. In this report, we present the third case of *ectopia cordis* in a twin pregnancy. We made the diagnosis of *ectopia cordis* as early as 9 weeks' gestation, which was 3 weeks after the fusion of the lateral folds in the thoracic area. The 3D reconstructed ultrasound, as well as color Doppler ultrasound, was also utilized to assist the diagnosis. We

also confirmed multiple fetal anomalies of Twin B at 20 weeks' gestation. In our case, we present the fetal *ectopia cordis* with a 3D high definition (HD) live rendering image. The HDlive technology uses an adjustable virtual light source, allowing the operator to create lighting and shadowing effects, thereby increasing depth perception [5]. Few research projects have demonstrated HDlive images as an innovative technique and a useful tool for a more realistic visualization of the embryo and the fetal anomalies [6].

We present here the earliest diagnosis of a twin fetus with *ectopia cordis* by fetal color Doppler and 3D reconstructed ultrasound.

Conflicts of interest

The authors have no conflicts of interest.

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