A Rare Case of Fetal Cardiac Hypertrophy Developing into Acute Circulatory Insufficiency and Fetal Compromise in Type 1 Diabetic Pregnancy

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Case Report

A Rare Case of Fetal Cardiac Hypertrophy Developing into Acute Circulatory Insufficiency and Fetal Compromise in Type 1 Diabetic Pregnancy

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Abstract

Fetal cardiac hypertrophy (CH) in pregnant women with diabetes is believed to be a benign condition. We encountered a rare case of fetal CH in a pregnant woman with type 1 diabetes, which developed into severe fetal circulatory insufficiency and acidemia. Fetal echocardiography at 37-week gestation showed cardiomegaly with a ventricular hypertrophy. Cardiac function was impaired, and pulsed Doppler findings indicated circulatory failure. The patient was diagnosed with fetal compromise due to fetal CH, and a large for gestational age boy was delivered by an urgent cesarean section. Despite myocardial hyperplasia and left ventricular outflow tract stenosis, the neonate was hemodynamically stabilized by fluid resuscitation alone. Although the neonatal course was favorable, we speculated that the neonate was on the verge of death because he was already acidemic at birth. Therefore, comprehensive fetal echocardiography should be performed in pregnant women with diabetes, and clinicians should not miss the optimal timing of delivery.

Keywords: Circulatory insufficiency, fetal cardiac hypertrophy, maternal diabetes

Introduction

Diabetes mellitus in pregnancy can cause adverse maternal and fetal effects. Fetal cardiac hypertrophy (CH) is a benign complication of diabetes mellitus in pregnancy. Although rare, several previous studies have reported CH cases resulting in stillbirth or neonatal death.[1,2] However, the clinical condition during fetal life for CH patients has not been fully understood because of the rarity of the condition. Further, it is unclear whether identifying the complication *in utero* can improve fetal prognosis. Herein, we report a case that was successfully managed by detecting hemodynamically deteriorated fetal CH in pregnant women with type 1 diabetes.

Case Report

The patient was a 35-year-old multiparous woman whose previous pregnancy had resulted in a cesarean delivery of a term neonate. She had developed type 1 diabetes at 27 years of age and was treated using a continuous insulin pump. In the present pregnancy, diabetes was continuously managed with insulin administration; however, hyperglycemia persisted until midgestation. Maternal serum glycated albumin and hemoglobin A1c levels were 21.4% and 7.0%, respectively, at 26-week gestation. She was referred to our hospital at 34-week gestation due to uncontrolled diabetes. The estimated fetal weight was 3284 g (±4.5 standard deviation), with normal amniotic fluid volume. The fetal heart seemed structurally normal without cardiomegaly or findings suggestive of a deteriorated circulatory state. Enhanced insulin therapy tended to ameliorate the patient's hyperglycemia in the third trimester. Maternal glycated albumin decreased to 15.7% at 34-week gestation [Figure 1]. On routine cardiotocography at 37-week gestation, the baseline fetal heart rate was 160 bpm with minimal variability, no acceleration, and mild late deceleration [Figure 2]. Fetal sonographic examination demonstrated cardiomegaly with a cardiothoracic area ratio

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of 43% and a remarkably hypertrophic ventricular wall. The interventricular septum (IVS) thickness was 19.5 mm (mean at 37-week gestation: 3.5 mm[3]). Diastolic compliance was expectedly reduced because of the thick ventricular wall and IVS. Tei index was 0.60 and 0.63 on the right and left, respectively (normal range; right 0.35 ± 0.05 and left 0.36 ± 0.06[4]), indicating impaired cardiac function. We noted A-wave reversal in the ductus venosus and a high preload index in the inferior vena cava. Further, we observed reversed end-diastolic velocity (REDV) of umbilical artery [Figure 3]. Consequently, we diagnosed the condition as fetal compromise caused by fetal CH with circulatory insufficiency. Suspecting neonatal heart failure, a cesarean section was performed in the presence of extracorporeal membrane oxygenation (ECMO) on stand-by. A male infant weighing 4718 g was delivered with 1-and 5-min Apgar scores of five and eight, respectively. The umbilical artery pH was 7.104. His cardiac contraction was preserved; however, myocardial hyperplasia and left ventricular outflow tract stenosis were observed on echocardiography [Figure 4]. Cardiomegaly was observed on a chest radiograph [Figure 5], with a brain natriuretic peptide (BNP) level of 3186.5 pg/mL (normally <18.4 pg/mL). Neonatal blood insulin level was 180.5 μU/mL (normal range, 2–5 μU/mL) and diagnosed as hyperinsulinemia. Despite the left ventricular outflow tract stenosis, the neonate was hemodynamically stabilized by fluid resuscitation alone and did not require the use of cardiovascular agents. Ventricular septal thickening and cardiomegaly gradually improved, and BNP level sharply decreased. He was discharged from the neonatal intensive care unit on day 23 after birth.

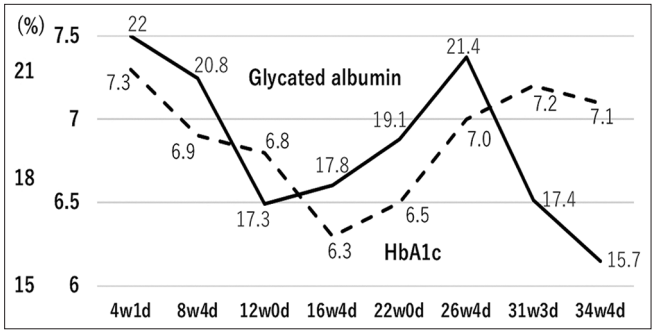

Figure 1: Changes in glycated hemoglobin and glycated albumin levels. Maternal serum glycated albumin and HbA1c levels were 21.4% and 7.0%, respectively, at 26 weeks. Glycated albumin decreased to 15.7% following enhanced insulin therapy

Discussion

CH is detected in 13%–44% of offspring delivered to diabetic mothers,[5] and it tends to occur in fetuses of type 1 diabetic mothers.[1] Although fetal CH due to maternal diabetes is believed to be a benign complication, some forms of fetal CH can be critical, considering that this finding is sometimes present in stillborn infants of diabetic mothers.[1,2] However, few studies have reported prenatal detection of CH with cardiac insufficiency using fetal sonography, and the clinical features of this disorder during fetal life have not been fully clarified. Furthermore, it is unclear whether prenatal diagnosis can improve fetal prognosis.

Studies have reported morphological and functional changes in the heart of fetuses of diabetic mothers (FDM). Morphologically, the ventricular wall, especially the IVS, is known to be hypertrophic in FDM,[6,7] which was the case in our patient. Functionally, a previous study described that the Tei index of the left ventricle is higher in FDM than that in fetuses of nondiabetic mothers and that a Tei index >0.39 is the optimal cut-off level for predicting adverse perinatal outcomes.[8] In the present case, the Tei index bilaterally increased to approximately 0.6, suggesting bilateral ventricular function impairment. Furthermore, it has been reported that diastolic dysfunction of both ventricles is a representative change in the heart of FDM.[9] Our grayscale findings indicated an apparent reduction in diastolic compliance due to myocardial hypertrophy. Pulsed Doppler findings of the ductus venosus and inferior vena cava indicated that the fetal heart could not deal with circulatory preload, probably due to diastolic dysfunction. Furthermore, we speculated that left ventricular stroke volume and cardiac output were reduced because REDV of umbilical artery was observed.

In our case, glycated albumin sharply decreased after 27-week gestation; however, the fetus developed CH thereafter. This suggests that fetal CH can occur even when maternal diabetes is well controlled. Furthermore, hyperinsulinemia, which is reported to cause CH,[5] was detected in the infant. Other cause of hyperinsulinism besides maternal diabetes, such as congenital hyperinsulinism, should be considered in the differential diagnosis because among the children with hypertrophic cardiomyopathy, the risk of death or heart transplantation is higher for those with inborn errors of metabolism.[10]

We assumed that the hemodynamic state of the affected fetus was severely impaired; therefore, we prepared for ECMO. Notably, however, the hemodynamic state was stabilized using

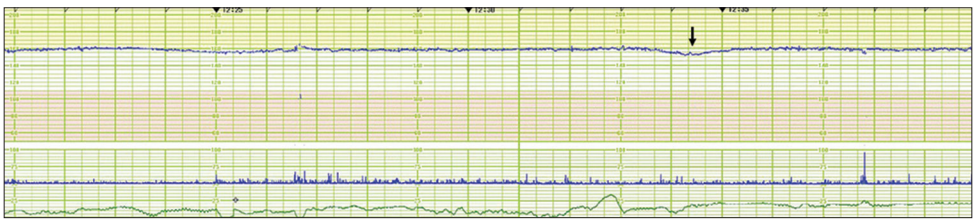

Figure 2: Fetal cardiotocograph. At baseline, the fetus presented with a heart rate of 160 bpm, minimal variability, no acceleration, and mild late deceleration (Arrow). The findings indicated fetal distress

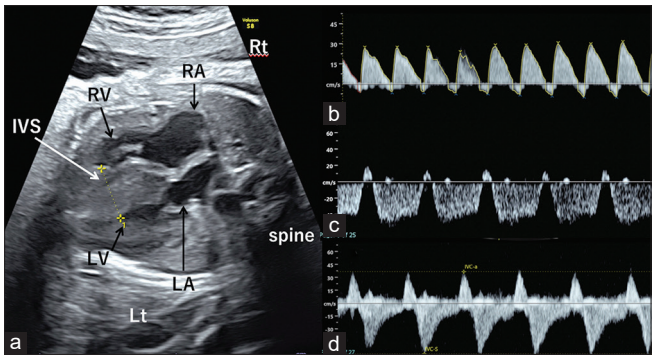

Figure 3: (a) Fetal heart ultrasound. (b) Reversed end-diastolic velocity in umbilical artery. (c) A wave reversal in ductus venosus. (d) Preload index in inferior vena cava. IVS: Interventricular septum, RV: Right ventricle, RA: Right atrium, LV: Left ventricle, LA: Left atrium

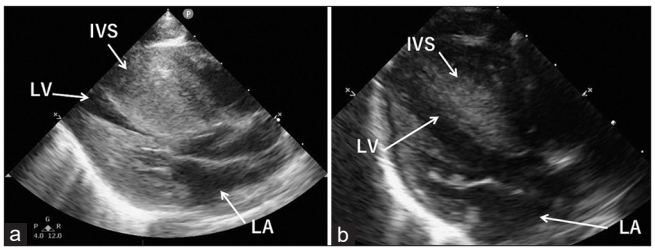

Figure 4: Echocardiograph of the neonatal heart. (a) Although cardiac contraction was adequate, myocardial hyperplasia and left ventricular outflow tract stenosis were observed on day 1. (b) Interventricular septal thickening was improved, and the left ventricular cavity was enlarged on day 19

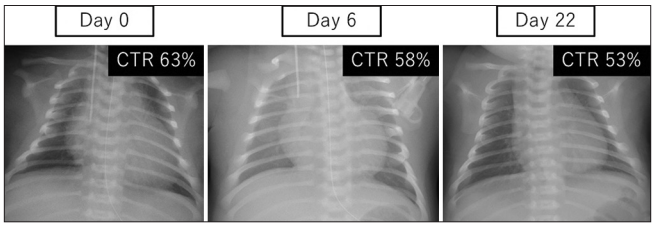

Figure 5: Chest radiograph of the neonate. Cardiomegaly was gradually ameliorated

volume expansion alone, and the CH spontaneously regressed. Although a favorable clinical course was obtained postnatally, we believe that the fetus presented in this case report was on the verge of death because he was already acidemic at birth.

In conclusion, our experience suggests that a careful structural and functional evaluation of the fetal heart in pregnant women with diabetes using detailed sonography should be performed to detect hemodynamically deteriorated CH, even if maternal glycemia is well controlled, and clinicians should not miss the optimal timing of delivery.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patients understands that her name and initial will not be published and due efforts will be made to conceal the identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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