

The case of a newborn manifesting heart failure caused by a chorioangioma

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Abstract

We reported the case of a newborn manifesting heart failure caused by a chorioangioma. A male infant was admitted to Kinki University Hospital Neonatal Intensive Care Unit (NICU) 4 hours after birth because of cyanosis. He had respiratory distress, and ultrasound echocardiography showed serious mitral regurgitation. An improvement of the mitral regurgitation 27 hours after birth resulted from olprinone hydrochloride administration, and mechanical ventilation was discontinued 2 days later. Pathologic examination of the placenta revealed a chorioangioma; a 290 g giant angioma adhering to a

250 g placenta, it therefore seemed likely that the cause of the respiratory distress was hemorrhagic pulmonary edema associated with the heart failure resulting from the blood-flow short circuit in the chorioangioma. Thus, in order to diagnose the cause of heart failure in a newborn infant, doctors should be aware of chorioangioma, and pathologic examination of the placenta could be important when an unusual course is observed in newborns.

Key words: infant, heart failure, placenta, hemangioma

Introduction

Respiratory distress is the major complaint of newborns admitted to neonatal intensive care units (NICU).¹ Causes of respiratory distress are not only limited to respiratory diseases, but could also involve cardiac and metabolic diseases to intestinal and nervous diseases; therefore, a differential diagnosis is important. Moreover, when cardiovascular diseases including heart failure are present, the diagnosis becomes difficult since these are often caused by diseases other than congenital cardiac diseases.² We reported the case of a newborn with hemorrhagic pulmonary edema caused by heart failure associated with a chorioangioma.

Case report

The patient was a male low-birth-weight infant. His mother was 33 years old, gravida 4,

para 4, with no specific events in her past pregnancies or deliveries. Polyhydramnion and fetal growth restriction were demonstrated by ultrasonography in the 29th week of gestation. Although cardiotocogram revealed a loss of variability in the fetal heart rate in the 38th week, aggressive medical intervention was not carried out. The boy was delivered by emergency caesarian section at 40 weeks and 2 days of gestation at a regional hospital because of fetal distress of unknown origin. Apgar scores were 9 and 9 at 1 and 5 minutes, respectively. Amniotic fluid was contaminated with meconium, while oronasal fluid was clear. He did not require resuscitation. His birth weight: 2400 g, height: 45.2 cm, and head circumference: 32.1 cm fulfilled the criteria for a small for dates infant. No specific abnormality was suspected for the placenta at that point. Thirty minutes after birth, he showed cyanosis with no significant signs of dyspnea. Since mitral regurgitation was

evident by echocardiography, dopamine hydrochloride was administered intravenously at a rate of 3.5 $\mu\text{g}/\text{kg}/\text{minute}$. He was then transferred to the NICU of Kinki University Hospital. During transfer, 40% oxygen was supplied, and oxygen saturation measured by a pulse oximeter (SpO_2) was kept over 90%.

He was admitted to the NICU 4 hours and 15 minutes after birth, his heart rate was 128 beats/minute, respiration rate was 77/minute, and he required 60% supplemental oxygen to maintain SpO_2 90%. Systolic murmurs, together with a galloping rhythm, were detected. Breathing sound was attenuated, and severe substernal and intercostal retractions were observed. The results of the blood examination were as follows: pH, 7.293; PCO_2 , 37.8 mmHg; PO_2 , 43.0 mmHg; HCO_3^- , 17.7 mmol/l; BE, -7.6; WBC, 20000/ μl ; Hgb, 15.6 g/dl; Hct, 50.4%; Platelet,

171000/ μl ; AST, 52 IU/l; CK, 74 IU/l; total protein, 5.5 g/dl; albumin, 3.2 g/dl; lactate, 3.3 mmol/l. Except for mild metabolic acidemia, values were normal for the newborn period. Although no congenital anomaly was demonstrated by ultrasound echocardiography, left atrial dilatation (Figure 1) and serious mitral regurgitation (Figure 2) were observed. Left ventricular diastolic dimension (21.1 mm) was estimated as 121.5% of the expected normal value for his body surface area. Ejection fraction was 68.2% and fractional shortening was 31.7%. As substernal and intercostal retractions were aggravated at 6 hours after birth, assisted ventilation was initiated. A large amount of tracheal fluid was discharged, which was serum-like light yellow, not meconium-like green. Chest x-ray after intubation showed markedly decreased

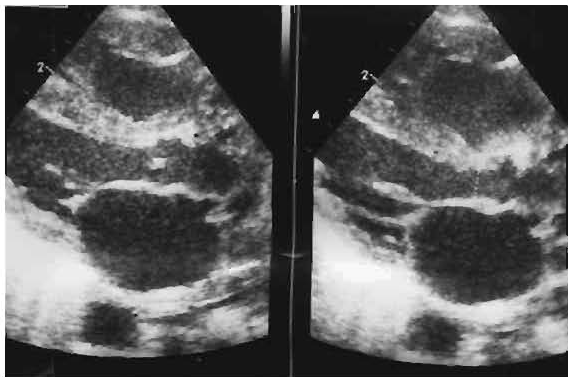


Fig. 1 Left atrial dilatation was shown on systolic (left) and diastolic (right) long axis views. Left ventricular diastolic dimension (21.1 mm) was estimated to be 121.5% of the expected normal value for his body surface area.



Fig. 3 Chest x-ray after intubation showed markedly decreased radiolucency in lung fields.



Fig. 2 Serious mitral regurgitation (↖) was revealed by color Doppler echocardiography (substernal view).



Fig. 4 Mitral regurgitation was not documented by color Doppler echocardiography 48 hours after birth (long axis view).

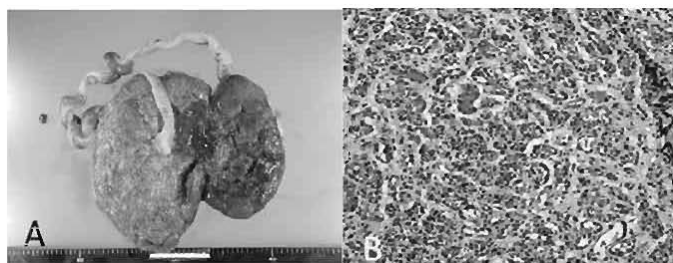


Fig. 5 A. A 290 g giant angioma (right) of approximately 15×8 cm adhering to a 250 g placenta (left)
B. Photomicrograph of the chorioangioma.
Intraparenchymal fibrous tissues and vegetabilization of the narrow vas capillary full of erythrocytes (hematoxylin-eosin staining, ×200).

radiolucency in lung fields (Figure 3); thus, olprinone hydrochloride injection at 0.2 $\mu\text{g}/\text{kg}/\text{minute}$ was initiated to reduce the afterload coupled with dopamine. On ultrasound echocardiography at 32 hours after birth, the reduction of both left atrial dilatation and mitral regurgitation was confirmed. Respiratory distress was also relieved and mechanical ventilation was discontinued 39 hours after birth. Subsequent ultrasound echocardiography did not reveal any abnormal findings (Figure 4), and olprinone hydrochloride and dopamine were discontinued 70 and 99 hours after birth, respectively. The boy was discharged from the NICU on the 15th day after birth, while the origin of heart failure was still unknown.

A few days after he was discharged, it was reported that pathologic examination of the placenta revealed a chorioangioma; a 290 g giant angioma of approximately 15×8 cm adhering to a 250 g placenta (Figure 5A). Consisting of several nodules, the angioma was covered with trophoblastic epithelium. Histologic examination demonstrated intraparenchymal fibrous tissues and excessive proliferation of blood vessels full of erythrocytes in chorionic villi; the findings suggested angioma³ (Figure 5B).

Discussion

Chorioangioma is a hamartoma derived from the primitive chorionic mesenchyme that is developed from angioblastic tissue.⁴ It is the most frequent nontrophoblastic tumor of the placenta with a frequency ranging from 0.01 to 1.3%.⁵ Fifty percent of all cases will cause maternal and fetal complications,⁶ and larger tumors have been more frequently associated with polyhydramnios, oligohydramnios, non-immune fetal hydrops, cardiomegaly with/with-

out heart failure, growth retardation, premature labor and intrauterine fetal death.³ Fetal complications arise through high output heart failure resulting from either anemia caused by intratumoral or fetomaternal hemorrhage or cardiac decompensation due to the shunting effect in the angioma.⁷

In this case, a giant angioma adhering to the placenta did not cause anemia, therefore, it was supposed that the origin of congenital heart failure was the increased cardiac load resulting from a cord venous-arterio vascular flow shunt in the angioma, namely, intrauterine high output cardiac failure. Polyhydramnion and fetal growth restriction found in the 29th week of gestation, loss of variability in fetal heart rate in the 38th week, and fetal distress just before delivery were also all related to intrauterine heart failure caused by the chorioangioma. Additionally, it is considered that the pathology of the respiratory distress was intraalveolar serum leakage causing increased pulmonary capillary pressure due to left-sided heart failure or hemorrhagic pulmonary edema. Increasing pulmonary blood flow after birth might have eventually caused hemorrhagic pulmonary edema 6 hours after birth.

Ultrasound and color Doppler flow mapping are important for the prenatal diagnosis of chorioangiomas, as an early prenatal diagnosis is crucial to minimize the risks to fetal well-being. Close surveillance of pregnancy and pregnancy termination by cesarean section at the earliest signs of fetal cardiac decompensation are indicated to reduce fetal and neonatal complications.⁸ Intrauterine treatments as the novel options include intravascular transfusion, fetoscopic devascularization, microcoil embolization, and intravascular injection of absolute alcohol.⁶ However, prenatal diagnosis and intrauterine

treatment may be limited, as it is likely that obstetricians rarely experience such conditions as chorioangiomas at routine checkups, while a postnatal primary diagnosis can be made relatively easily by visual examination of the placenta. The placenta should be examined to find abnormal lesions such as chorioangioma for a differential diagnosis if there is any unusual fetal and neonatal history, such as intrauterine growth restriction of unknown etiology, congenital heart failure, neonatal disseminated intravascular coagulation, or fetal or neonatal death.

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