

CASE REPORT

Successful Management of Fetal Hydrothorax associated with Hydrops

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ABSTRACT

We report a case of massive unilateral hydrothorax diagnosed at 18 weeks' gestation in a fetus with normal karyotype. The fetus was affected severely by hydrops from 22 weeks. Initially, we performed a pleurodesis, but due to worsening hydrothorax evolving into hydrops, we proceeded with insertion of a transplacental thoracoamniotic shunt. Improvement was evident from 1 week after the procedure followed by resolution of hydrothorax for the remainder of the pregnancy. After a cesarean delivery at 37 weeks, the neonate required prolonged neonatal intensive care unit stay. He was discharged when he was 2 months old and has remained stable until the present time.

Keywords: Fetal hydrothorax, Hydrops, Pleural effusion, Prenatal treatment, Thoracoamniotic shunt.

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INTRODUCTION

Fetal congenital hydrothorax or pleural effusion is a rare condition.^{1,2} While mild effusions remain stable or resolve, large effusions are associated with mass effect leading to mediastinal shift, pulmonary hypoplasia, hydrops, and/or abnormal cardiac function or arrhythmias.²⁻⁴ Fetuses presenting with hydrops and polyhydramnios due to large pleural effusion are at risk for preterm delivery and perinatal death due to pulmonary hypoplasia or heart failure.⁵

We present a case of unilateral large pleural effusion associated with hydrops in a fetus with anterior placenta

successfully managed with transplacental thoracoamniotic shunting.

CASE REPORT

A healthy 29-year-old primigravida was referred to our center at 18 weeks due to fetal unilateral hydrothorax. Her pregnancy course had been uncomplicated before diagnosis. At 19 weeks, we confirmed a large left pleural effusion with right mediastinal shift; we did not note other structural malformations. We performed an amniocentesis with a 22-gauge needle. After collecting amniotic fluid, we performed a thoracentesis retrieving 26 mL of yellow-tinged fluid. The pleural fluid showed no bacteria by Gram staining, leukocyte 300/ μ L with 77% lymphocytes, protein 1.63 gm/dL, and cholesterol 31 mg/dL. Chromosomal karyotype was reported as normal (male fetus). Maternal serology showed no acute toxoplasma, cytomegalovirus or herpes infection, and was negative for indirect Coombs and Venereal Disease Research Laboratory test. At 22 weeks, a repeat ultrasound showed massive hydrothorax, subcutaneous edema, and ascites (fetal hydrops) along with polyhydramnios (Fig. 1).

Due to the anterior location of the placenta, we deferred thoracoamniotic shunting and opted for pleurodesis carried out at 23 weeks: After aspirating 100 mL of pleural fluid, we injected 15 mL of fresh maternal blood into the pleural cavity under sonographic guidance using a 20-gauge needle. One week later, we

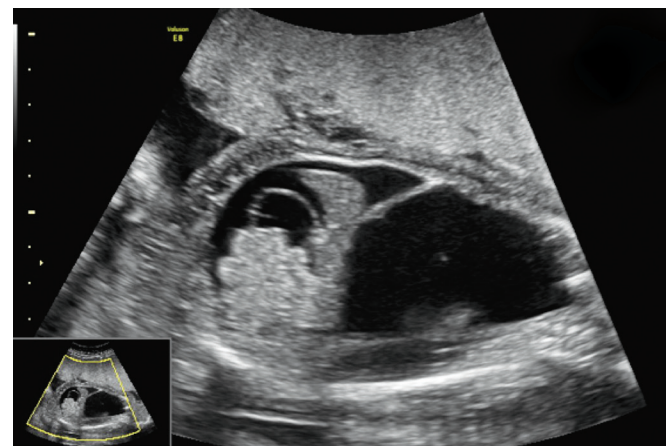


Fig. 1: Preoperative ultrasound image at 22 weeks' gestation showing massive hydrothorax with subcutaneous edema and ascites (hydrops)

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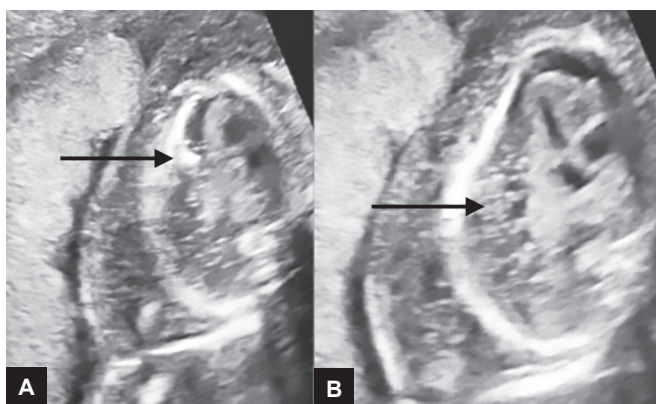


Fig. 2A and B: Pleurodesis at 23 weeks' gestation. The picture on the left shows the echogenic tip (arrow) of the needle entering the fetal thorax. The picture on the right shows observed blood regurgitation (arrow)

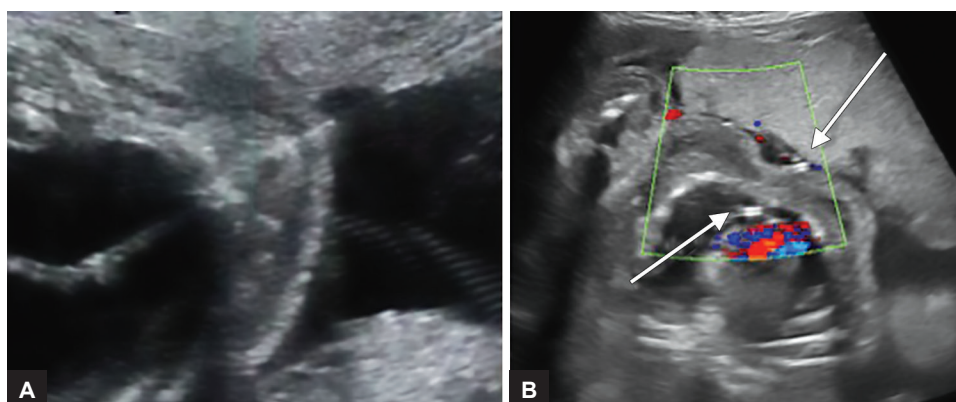
noted accumulation of pleural fluid with worsening hydrothorax (Fig. 2).

At 24 weeks, the fetus became severely hydropic and showed intermittent episodes of bradycardia. After reviewing the risks and benefits with the patient, we

obtained consent to perform a transplacental fetal thoracoamniotic shunting using a silicone double-pigtail catheter inserted through a 3 mm introducer (Harrison shunt, Cook Medical, Inc., Spencer, IN, USA) placed in the left pleural space under ultrasound guidance (Fig. 3). The procedure was uncomplicated. One week later, the fetal hydrothorax had decreased considerably. At 33 weeks, there was only residual laminar left hydrothorax while ascites and subcutaneous edema had resolved (Fig. 4).

After administering late preterm antenatal corticosteroids, we delivered a 2,950-gm newborn at 37 weeks via cesarean. Apgar scores were 9 at 1 and 5 minutes. During delivery, the pigtail catheter was displaced inadvertently from the fetal thorax before it could be clamped. This led to neonatal pneumothorax requiring mechanical ventilation. The newborn developed pulmonary hypertension that resolved with medical management. After 6 weeks of hospitalization, he was discharged in stable condition.

At the present time, the infant remains stable achieving normal psychomotor and anthropometric milestones (Fig. 5).



Figs 3A and B: On the left picture, we observe the needle inside fetal thorax after crossing the placenta. On the right, the white arrows point to both ends of the pigtail catheter: One inside the thorax and the other in the amniotic cavity

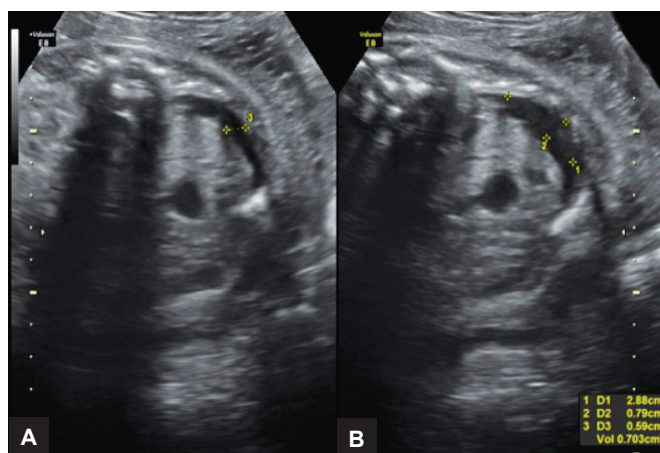


Fig. 4: At 32 weeks' gestation, the fetal thorax shows residual laminar effusion in the left thorax posttransplacental thoracoamniotic shunting



Fig. 5: Newborn at 2 months in stable condition after hospital discharge

DISCUSSION

Fetal hydrothorax refers to pleural effusion, which can be either primary or secondary. Its incidence ranges from 1:10,000 to 1:15,000 pregnancies.¹ Primary effusions are due to lymphatic leakage resulting in raised intrathoracic pressure that may progress to hydrops.⁶ The most common form of primary pleural effusion is chylothorax. Chylothorax is diagnosed by cytological and biochemical analysis of the fluid aspirate: Yellow-colored, with lymphocyte count >70–80%, and fluid protein and cholesterol lower than their serum counterparts.^{2,3} The prognosis depends largely on the underlying cause. Perinatal outcomes are worsened by associated malformations (as much as 25%) and aneuploidy (7–12% of all cases).^{7,8} The majority of cases are mild,⁹ but severe cases ensue when the fluid expands over >50% of the thorax. Severe pleural effusion is associated with elevated perinatal morbidity and mortality which are much higher in the presence of fetal hydrops.¹⁰ Hydrops in a fetus with primary hydrothorax carries a poor prognosis, and is an independent predictor of poor outcomes with perinatal mortality rate as high as 50%.¹⁸

In our case, the fetal pleural effusion evolved into severe hydrops with episodes of bradycardia. Initially, we managed this case with thoracentesis; however, it did not succeed and the hydrothorax progressed into hydrops. Next, due to the anterior placental location, we performed a pleurodesis, instead of shunting, injecting maternal blood. Pleurodesis was first described in 2001 by Okawa et al¹¹ with good outcomes using OK-432 as the sclerosant agent. Because OK-432 is not free of potential risks,¹² other clinicians have used maternal blood instead. The first report of pleurodesis with intrapleural injection of maternal blood was reported by Parra et al¹³ with also good results and less fetal risks. Nonetheless, in our patient, pleurodesis using maternal blood was not successful either and the fetus presented with worsening hydrops and new onset of bradycardia episodes. Next, we opted for transplacental thoracoamniotic shunting performed without any complications leading to resolution of pleural effusion and hydrops and normalization of the fetal heart rate. This technique was described by Seeds and Bowes¹⁴ for the first time in 1986. Thoracoamniotic shunting is the gold standard therapy for pleural effusion in the setting of hydrops, with a survival rate of 33 to 66%. Rates of success are higher with the use of a double-pigtail catheter.¹⁵ After shunting placement, close monitoring of the fetus by ultrasound scans is warranted. Although *in utero* displacement of the catheter has been reported in as much as 23% of cases,² we did not encounter that problem. Rather, it was at delivery that the catheter dislocated leading to neonatal pneumothorax.

The early response to shunting seen in our case was pivotal because it allowed fetal lung expansion

(preventing pulmonary hypoplasia) and restoration of the displaced mediastinal anatomy. We suggest that in cases of diagnosis in the second trimester and large effusions with hydrops, the most appropriate treatment is thoracoamniotic shunt even with an anterior placenta. The mode of delivery can be based on obstetric criteria. Delivery should occur at a tertiary care center with good neonatal support where potentially an ill neonate can have access to an intensive care unit, especially if ventilation support is required as in this case.

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