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Case Report

In Utero Intervention for Isolated Fetal Pleural Effusion: A Case Report

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ABSTRACT

Fetal pleural effusion is a rare condition easily diagnosed by antenatal ultrasound. It can be classified into two categories: primary and secondary. Primary pleural effusion is mainly attributed to defective lymphatic drainage in the fetus. Secondary pleural effusion can be caused by conditions that affect the fetal cardiac function, fetal anemia, fetal infections, chromosomal aberrations, genetic diseases, or congenital malformations that disrupt the lungs and mediastinal architecture. The clinical course is variable depending on the severity, underlying cause, gestational age at diagnosis, and the presence or absence of other congenital abnormalities. We present a case of isolated fetal pleural effusion where in utero therapy included thoracocentesis followed by the insertion of a thoracoamniotic shunt.

Keywords: in utero therapy, fetal pleural effusion, fetal shunt, intervention

INTRODUCTION

Fetal pleural effusion is a rare condition that can be easy to diagnose with routine ultrasound assessment. Its incidence is approximately 1 in 10,000–15,000 pregnancies.[1,2] Although it is easy to diagnose, management may prove to be challenging. Fetal pleural effusion can be classified into two distinct categories: primary and secondary. Primary pleural effusion can be attributed mainly to defective lymphatic drainage in the fetus. Secondary pleural effusion can be caused by conditions that affect the fetal cardiac function, fetal anemia, fetal infections, chromosomal aberrations, genetic diseases, or congenital malformations that disrupt the lungs and mediastinal architecture.[3–6]

The clinical course is highly variable and depends on the severity, underlying cause, gestational age at diagnosis, and other congenital abnormalities. Mild isolated pleural effusion can have a benign course that does not necessitate prenatal intervention. More severe cases have more controversial management options, which include thoracocentesis, thoracoamniotic shunting, and pleurodesis.[7,8] Thoracocentesis, although less invasive than thoracoamniotic shunting, may prove to be fruitless due to the quick reaccumulation of the fluids in the pleural space. For this reason, thoracoamniotic shunting has been proposed as a reasonable option if reaccumulation is rapid.[4,9]

CASE REPORT

We present a 43-year-old woman, gravida 6, para 3, and with two previous miscarriages. The patient granted permission to publish her case details and ultrasound images. All her previous deliveries were full-term spontaneous vaginal deliveries. She is not known to have any chronic illnesses. She was infected with COVID-19 in the second trimester of the current pregnancy; however, because of the lack of evidence correlating COVID-19 infection and fetal pleural effusion, we were not able to discern any causality. She was referred from the Sultanate of Oman at 27 + 1 weeks' gestation with the diagnosis of fetal pleural effusion. She underwent initial assessment with a late anomaly screen, fetal echocardiography, and TORCH (toxoplasmosis, others, rubella, cytomegalovirus, herpes infections) screen using maternal blood. We confirmed isolated unilateral pleural effusion, and no other fetal structural abnormalities were seen (see Fig. 1). All blood work including TORCH screen were negative. Fetal echocardiography showed normal fetal heart structure and no evidence of cardiomyopathy. Middle cerebral artery Doppler revealed normal peak systolic

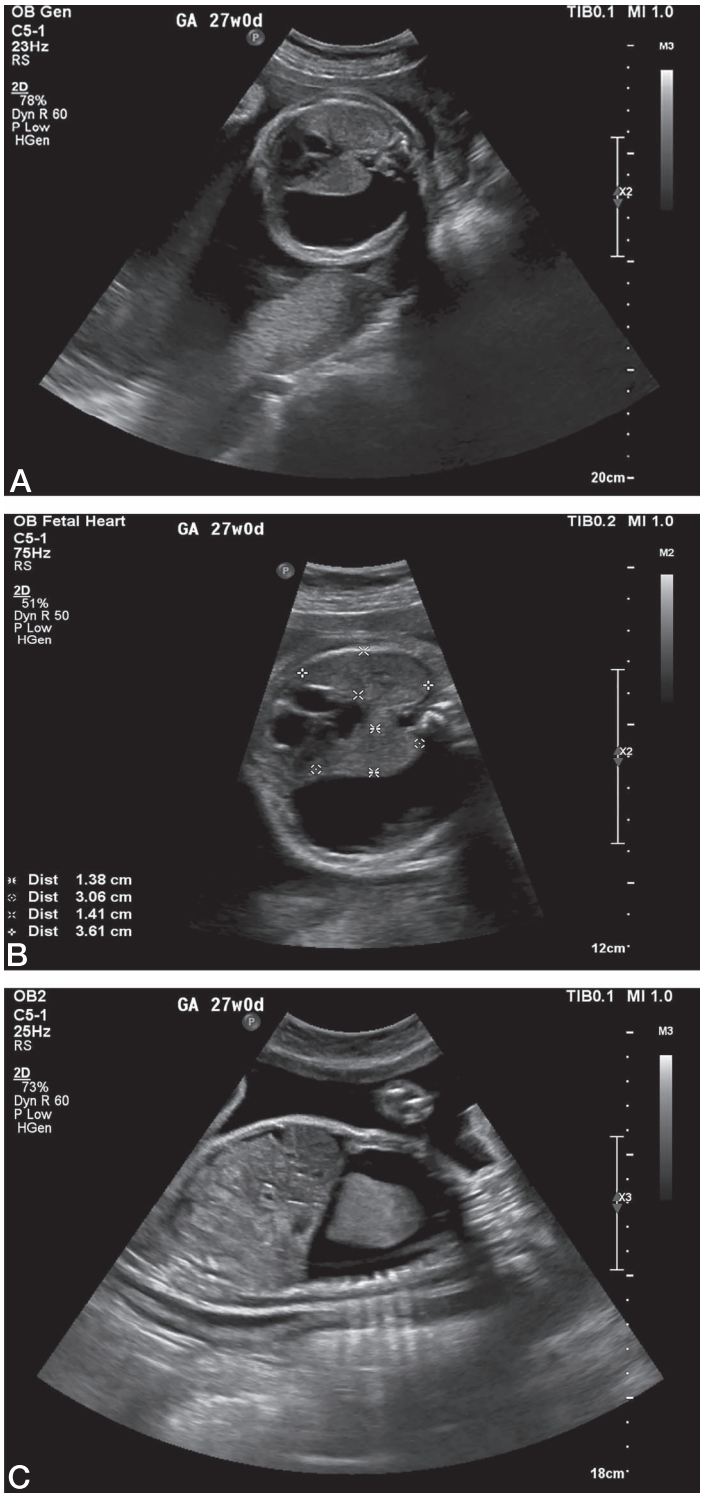

Figure 1. Ultrasound images obtained prior to shunting. (A) Pleural effusion seen. (B) Small ipsilateral lung, heart shift. (C) Sagittal view for the chest pleural effusion seen. GA: gestational age.

velocity for the gestational age, thus excluding any risk of fetal anemia.

We proceeded with amniocentesis using fluorescent in situ hybridization to check the fetal karyotype, which showed normal number of chromosomes 13, 18, 21 X and Y. Thoracocentesis was done initially in the hope that the pleural effusion would not reaccumulate. We aspirated 70 mL of the fluid and sent it for analysis. It predominantly showed chronic inflammation, giving a higher suspicion for the diagnoses of primary isolated chylothorax. At follow-up 5 days later showed reaccumulation of the pleural effusion and mild ascites. We discussed the next management option of thoracoamniotic shunting. Risks and benefits were discussed with the couple, and they agreed to proceed with the procedure.

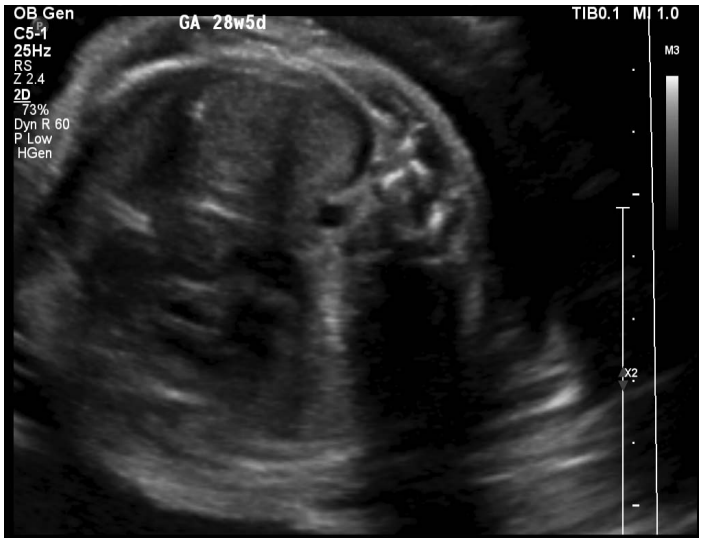

Figure 2. Ultrasound images obtained 5 days after shunting. Expansion of the lungs and resolution of the pleural effusion seen. GA: gestational age.

On the day of the procedure, the patient was requested to present to the prenatal procedure room having fasted for at least 6 hours. After signing an informed consent, she was given 75 mg of intramuscular Pethidine for pain relief. Lidocaine 2% was injected at the proposed insertion site of the trocar. We used a Rocket Medical KCH drain, double pigtail tubing that was introduced using a stainless-steel trocar with an outside diameter of 1.65 mm. Insertion was easy; using the introducer, one end of the tube was dislodged into the pleural space, and the other end was released outside the fetal chest and into the amniotic cavity. Both ends were seen clearly by ultrasound; the outside end was seen just outside the fetal chest skin. After the double pigtail drain was inserted under ultrasound guidance, there was still fluid seen in the pleural space. At that point, we were not sure if the shunt was draining properly. We therefore proceeded with a second thoracocentesis and removed another 70 mL of fluid in the hope that the pressure of the expanded lung would allow further drainage from the shunt. The patient was observed for 6 hours post procedure. Fetal heart activity was recorded using nonstress testing. The patient was discharged in a stable condition and followed up in another 5 days. The ultrasound assessment done after 5 days showed complete resolution of the ascites and marked reduction in the pleural effusion (see Fig. 2).

The patient wanted to continue follow-up in her local hospital in Oman. Detailed reports were given to the patient to present to her managing physician. Five days after the procedure she experienced labor like pain and reported to her local hospital. She was observed for a couple of days, and another ultrasound was done that showed reaccumulation of the fluid in the pleural cavity; therefore, the possibility of blockage of the tube was entertained. The patient progressed to preterm labor and eventually was delivered via cesarean delivery. After delivery, the shunt was removed, and bilateral chest tubes were inserted. Other than the reaccumulation of the pleural effusion, there were no gross abnormalities detected postnatally.

DISCUSSION

Thoracoamniotic shunting was first introduced in 1986 by Seeds et al.[10] With the advancement of ultrasonography and medical equipment, the procedure became more frequent worldwide. Although it has become more widely adopted, it can still lead to various complications, such as preterm labor, premature preterm rupture of membranes, placental abruption, infections, maternal mirror syndrome in cases of fetal hydrops, and fetal death.[4,5,11] Other complications related to the shunt itself include dislodgement into the thoracic cavity and blockage, occasionally necessitating a repeat procedure.[11] In one of the largest reported series conducted in Toronto, Canada, in 2021 by Kelly et al,[12] which included 132 fetuses, the overall survival rate was 65%. In this report, it was concluded that gestational age was the only independent predictor of survival and normal neurodevelopmental outcome. Although thoracoamniotic shunting remains a relatively new procedure with variable outcome, it has been shown to be an attractive treatment option for significant fetal hydrothorax.

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