

Research Letter

Treatment of unilateral fetal pleural effusion by intrauterine thoracocentesis

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Fetal pleural effusion refers to an accumulation of fluid in the pleural space. It is most commonly attributed postnatally to chylothorax, a primary lymphatic abnormality. Following the diagnosis of pleural effusion, detailed investigation is essential to exclude other conditions (secondary pleural effusion). The natural history of the lesion is variable. The effusion may resolve spontaneously, remain stable or progress to hydrops and fetal or neonatal demise [1]. The treatment modalities include thoracocentesis, pleuro-amniotic shunting and pleurodesis. The optimal antenatal management is controversial [2,3]. We report a case of isolated pleural effusion diagnosed at 32 weeks of gestation that we successfully treated with thoracocentesis, resulting in a good outcome.

The patient was a 30-year-old woman, G2P1A0, referred to our hospital for further management of fetal right pleural effusion at 32 weeks' gestation. According to the referral information, the pregnancy had been unremarkable until 32 weeks' gestation. She had not had any illness or recent infectious disease, had not been exposed to teratogenic agents during the pregnancy, and did not have a family history of congenital anomalies. The Down syndrome screen during her second trimester was negative. She had a history of previous cesarean section as a result of preeclampsia and fetal distress.

Sonography demonstrated fetal right pleural effusion (Fig. 1). The right lung was compressed and displaced slightly to the midline. There was no evidence of pericardial effusion or gross cardiac defect. There was no other fetal anomaly detectable during the detailed ultrasound examination. One week after the initial visit to our hospital, an increase in the volume of the fetal right pleural effusion occurred. Trans-abdominal fetal thoracocentesis at 33 2/7 weeks' gestation was suggested and the patient consented to undergo the procedure. Using a 22-gauge needle and sonographic guidance, 70 mL yellow fluid was removed from the right fetal

thorax. The fluid accumulation recurred within 48 hours. Repeated ultrasound scans over the course of the next few days showed an increasing volume of pleural fluid accumulation, with a mediastinal shift of the right lung. At 34 weeks' gestation, pericardial effusion, cardiomegaly and subcutaneous edema occurred. Hydrops was suspected and the mediastinal shift of the right lung progressed. Prompt delivery of the fetus was suggested. Thoracocentesis was performed prior to the cesarean delivery and 150 mL of pleural fluid was removed. A female infant weighting 2300 g was delivered, with Apgar scores of 4 and 8 at 1 and 5 minutes, respectively.

The infant was born in relatively good condition. She breathed spontaneously and was transferred to the neonatal intensive care unit. Nonetheless, because of cyanosis with chest retraction, she required intubation and ventilation shortly after birth. A chest X-ray revealed massive right pleural effusion. A chest tube was inserted a few hours after delivery. Biochemical analyses of the pleural fluid (triglycerides: 747 mg/dL; lymphocyte proportion: 92%) confirmed the diagnosis of chylothorax. The infant was extubated on Day 3. The pleural effusion volume decreased gradually. The chest tube was removed on Day 13. The infant received conservative management and was discharged 28 days after birth. Subsequent follow-up showed normal growth and development.

Fetal pleural effusion is a rare condition. The incidence is estimated to be 1/10,000 to 1/15,000 pregnancies [4]. Primary pleural effusions are caused by lymphatic leakage and can be unilateral or bilateral [3]. In a recent study, the survival rate of primary pleural effusion without intrauterine treatment was 59% (35% with hydrops and 73% without hydrops) [2]. The optimal management of fetal pleural effusion in the prenatal period is still a matter of debate.

Observation is appropriate in the case of primary, small, nonhydropic effusions, because spontaneous regression can occur [1,3]. Klam et al. suggested that conservative management is the preferred management option in most cases of primary fetal hydrothorax [5]. However, Knox et al. showed that prenatal intrauterine drainage did not improve perinatal survival compared with no drainage, except in fetuses with

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Fig. 1. Sonogram of fetus at 32 weeks of gestation showing right pleural effusion.

hydrops [6]. If effusions enlarge rapidly, or hydrops develops, prenatal intervention should be offered [2]. Recent research supports the notion that *in utero* intervention improves the survival of fetuses with persistent effusions and hydrops [2,7].

The treatment options include repeated thoracocentesis, thoraco-amniotic shunting, and pleurodesis. Although there are no statistically significant differences between the treatments [2,7], thoraco-amniotic shunting appears superior to other treatments [2,3]. Thoraco-amniotic shunting is a more invasive procedure and may not be available in every hospital. Catheter migration, obstruction, preterm premature rupture of amniotic membrane, and chorioamnionitis are the main complications [3]. Pleurodesis is a new form of prenatal treatment of fetal hydrothorax, which is achieved by intrapleural injection of OK-432, a sclerosing agent. Data on this new procedure are limited [2,3]. Further studies are needed to clarify the role of this therapy for treatment of fetal hydrothorax.

Pleural effusions sometimes resolve *in utero* after thoracocentesis [8,9]. However, recurrence of the pleural effusion occurs in the majority of cases [1,5,7]. If recurrence of pleural fluid accumulation ensues, repeat thoracocentesis may be considered. However, repeat thoracocentesis could cause hypoproteinemia, which favors the development of hydrops [1,3]. Thoracocentesis may be performed before delivery and seems to facilitate neonatal resuscitation [10]. It allows newborns to breathe spontaneously, be more easily ventilated, and reduces emergency chest tube insertion [11].

Premature delivery and the presence of hydrops consistently are regarded as the most important poor prognostic indicators of fetal hydrothorax [3]. Gestational age is an

important factor in deciding treatment [7]. Aubard et al. suggested that before 32 weeks of gestation, the greatest fetal survival can be obtained by placement of a pleuro-amniotic shunt. Between 32 and 37 weeks, thoracocentesis provides the best results [1].

Primary fetal pleural effusion is a rare pathology in which antenatal therapeutic approaches range from simply observing to performing highly invasive *in utero* interventions. From our experience in treating this case and researching the literature, we decided to proceed conservatively at first, with close follow-up. Then, if intervention was necessary, thoracocentesis should be the initial treatment modality of fetal pleural effusion because resolution could occur. If the pleural effusion progressed quickly or hydrops developed, the next step of treatment should be based on the gestational age of the fetus. When the gestational age is 32–34 weeks, prompt delivery should be suggested. When the gestational age is less than 32 weeks, thoraco-amniotic shunting should be considered. We also recommended thoracocentesis prior to delivery to facilitate neonatal resuscitation.

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