

Case Report

Case of Thoracoamniotic Shunt Placement for Treatment of Pleural Effusion in the Setting of Congenital Diaphragmatic Hernia Complicated by Hydrops

Antonio Santos¹, Kelsey Montgomery², David Kays³ and Tiffany Tonismae¹

¹Department of Obstetrics and Gynecology, Bayfront Health Hospital, USA

²Department of Obstetrics and Gynecology, St Vincent Health, USA

³Department of Obstetrics and Gynecology, John Hopkins All Children's Hospital, USA

Abstract

To review a case of novel in utero thoracoamniotic shunt placement for pleural effusion in the setting of congenital diaphragmatic hernia (CDH).

Case Presentation

A 37 yo G1P0 with known diagnosis of right CDH on growth scan at 20+1 weeks GA with moderate pleural effusion was evaluated for fetoscopic Endoluminal Tracheal Occlusion (FETO) candidacy at 24+2 weeks GA. The US demonstrated normal growth with right CDH containing liver and gallbladder with moderate right pleural effusion. ECHO obtained with exaggerated levoposition due to right pleural effusion but otherwise normal structure and function. Thoracoamniotic shunt placement was recommended at that time but the patient declined due to concern for preterm delivery with a poor outcome. Repeat evaluation at 26+2 weeks GA demonstrated similar findings of normal growth and large pleural effusion with new small ascites. Worsening polyhydramnios with preterm contractions warranted urgent repeat evaluation at 29+3 weeks GA, and US demonstrated concern for hydrops fetalis with severe polyhydramnios, skin edema, large abdominal ascites, and worsened large pleural effusion. Patient underwent placement of a singular thoracoamniotic shunt at 29+4 weeks gestation and was hospitalized on tocolytics for five days post-procedure. Amniocentesis was obtained at that time and demonstrated a normal male karyotype of 46, XY. Following placement of the shunt, the patient was transferred under our care at 30+5 weeks GA for the remainder of pregnancy. Evaluation of the fetus was performed including fetal echocardiogram, serial ultrasounds with Maternal Fetal Medicine (MFM), and

consultation with the CDH team at Johns Hopkins All Children's Hospital (JHACH). Imaging at 34 weeks was consistent with a right-sided CDH containing bowel, liver, and gallbladder. There was no evidence of residual pleural effusion or ascites and the thoracoamniotic shunt remained in place for the duration of pregnancy. The CDH was categorized as moderate to less severe based on MRI with a 95% likelihood of survival and 5-30% ECMO risk based on protocolized treatment at JHACH. Patient was monitored regularly by ultrasound through the MFM office with planned delivery via scheduled cesarean at 37+3 weeks gestation. Per CDH protocol, two doses of antenatal corticosteroids were administered prior to delivery. The male infant was delivered via primary low transverse cesarean section with Apgar scores of 7 and 8 at 1 and 5 minutes respectively. Birthweight was noted to be 2770 g. Immediately following delivery, the infant was evaluated by the CDH team with resuscitation including endotracheal intubation and positive pressure ventilation in the operating room prior to being taken to the CDH Intensive Care Unit (ICU). Neonate was monitored in the ICU and taken for primary surgical repair of CDH on day of life (DOL) 3 without need for patch placement. He was continued on bowel rest with Total Parenteral Nutrition (TPN) for 12 days following surgery. He was extubated to Continuous Positive Airway Pressure (CPAP) on DOL 9 and transitioned to nasal cannula at 0.4 L on DOL 15. He initiated breastfeeding on CPAP and began small volume oral feeds with breast milk and fortified formula on nasal cannula while continuing TPN for nutrition. TPN was discontinued on DOL 35. The neonate was discharged home on DOL 49 on room air, tolerating oral feeds without difficulty. Neonate continued close follow-up with the CDH team following discharge.

Discussion

CDH is a rare birth defect occurring in approximately 1 in 2500-3000 live births of which right sided occurs only 10-15% of cases. The defect requires care of a multidisciplinary team with the option of in utero tracheal occlusion for select candidates and the remainder having repair following delivery. While there is limited data about risks and management of pleural effusion in the neonate, there is very limited data for in utero management options. While fetoscopic

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***Corresponding author:** Tiffany Tonismae, Department of Obstetrics and Gynecology, Bayfront Health Hospital, USA, Tiffany.Tonismae@orlandohealth.com; TRTonismae@gmail.com

Endoluminal Tracheal Occlusion (FETO) procedures are performed for a select few candidates, there is concern for worsening pulmonary hypoplasia in patients that do not undergo fetal surgery, and with additional pleural effusion, concern for additional pulmonary compromise and growth constriction. This case demonstrates the use of thoracoamniotic shunt placement to manage pleural effusion in the

setting of fetal hydrops. While it is unknown if the outcome would be similar without shunt placement, precedence of prior cases of pleural effusion in the setting of other thoracic malformations suggest that pleural effusion may represent a worsening prognosis without correction. Additional cases are needed to compare shunt placement to conservative treatment to determine a change in prognosis, if any.