CLINICAL STUDY

Percutaneous In Utero Thoracoamniotic Shunt Creation for Fetal Thoracic Abnormalities Leading to Nonimmune Hydrops

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ABSTRACT

Purpose: To describe a transabdominal, transuterine Seldinger-based percutaneous approach to create a shunt for treatment of fetal thoracic abnormalities.

Materials and Methods: Five fetuses presented with nonimmune fetal hydrops secondary to fetal thoracic abnormalities causing severe mass effect. Under direct ultrasound guidance, an 18-gauge needle was used to access the malformation. Through a peel-away sheath, a customized pediatric transplant 4.5-F double J ureteral stent was advanced; the leading loop was placed in the fetal thorax, and the trailing end was left outside the fetal thorax within the amniotic cavity.

Results: Seven thoracoamniotic shunts were successfully placed in five fetuses; one shunt was immediately replaced because of displacement during the procedure, and another shunt was not functioning at follow-up requiring insertion of a second shunt. All fetuses had successful decompression of the thoracic malformation, allowing lung reexpansion and resolution of hydrops. Three of five mothers had meaningful ($>$ 7 d) prolongation of their pregnancies. All pregnancies were maintained to $>$ 30 weeks (range, 30 weeks 1 d–37 weeks 2 d). There were no maternal complications.

Conclusions: A Seldinger-based percutaneous approach to draining fetal thoracic abnormalities is feasible and can allow for prolongation of pregnancy and antenatal lung development and ultimately result in fetal survival.

ABBREVIATIONS

AFI = amniotic fluid index, CPAM = congenital pulmonary airway malformation, MFM = maternal-fetal medicine

Since first described in 1988, thoracoamniotic shunt placement has been a well-established treatment for thoracic fetal abnormalities, including fetal pleural effusions, chylothoraces, and type I congenital pulmonary airway malformations (CPAMs) (1). Mortality is usually due to fetal nonimmune hydrops, a severe condition in which excessive fluid accumulates in fetal soft tissue and serous cavities (Fig 1) (2). In the absence of maternal circulating red cell antigens and fetal anemia, this condition is called nonimmune hydrops. There are numerous causes of nonimmune hydrops, but it is often seen in cases of intrathoracic masses, secondary to mediastinal shift, decreased venous return, and severely depressed cardiac output (3). Long-term compression of the normal fetal lung tissue can lead to pulmonary hypoplasia and severe neonatal morbidity and death (4). The indication for antenatal intervention is the presence of an isolated large structural abnormality that causes nonimmune fetal hydrops in an extremely premature fetus

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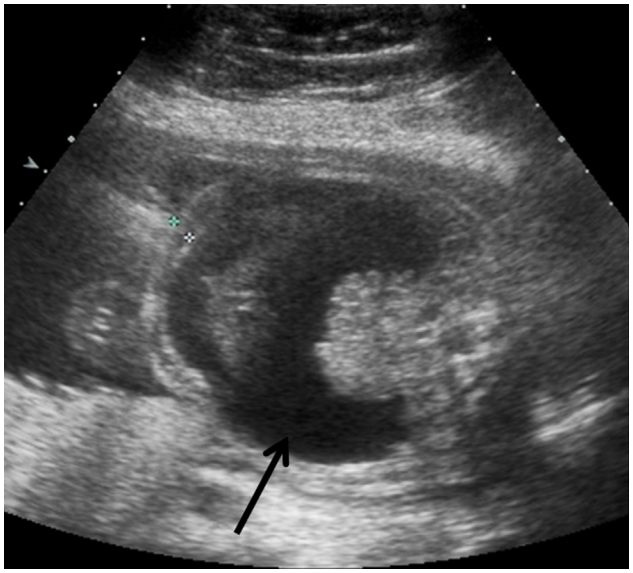

Figure 1. Fetal hydrops. Transverse view of the fetal abdomen obtained as part of a fetal ultrasound scan. There is a large volume of ascites (arrow) and skin thickening (calipers), which is consistent with nonimmune fetal hydrops. (Available in color online at *www.jvir.org*.)

before 30–32 weeks' gestational age (5–7). The goal of this therapy is to reverse the hydrops, relieve lung compression, and prolong the pregnancy, avoiding fetal death, reducing the risks of prematurity, and allowing for definitive neonatal treatment (8).

Traditionally, thoracoamniotic shunts were created using the trocar technique, which is performed by direct puncture of the fluid collection using a central stylet within a catheter (9). This article describes an ultrasound-guided, transuterine Seldinger-based percutaneous approach to create a shunt for treatment of fetal thoracic abnormalities as performed by a maternal-fetal medicine (MFM) and interventional radiology (IR) team.

MATERIALS AND METHODS

The institutional review board approved this retrospective study and waived informed consent. This study was compliant with the Health Insurance Portability and Accountability Act. In addition, a special committee of the institutional review board was convened in each case to grant approval for compassionate treatment. Five consecutive patients underwent creation of a thoracoamniotic shunt between December 12, 2007, and July 7, 2012 (the date of institutional review board submission).

The fetuses presented with nonimmune fetal hydrops secondary to fetal thoracic abnormalities causing severe mass effect. All of the fetuses underwent detailed ultrasound evaluation to confirm the diagnosis of fetal thoracic abnormality and exclude other anomalies. Amniocentesis was performed to evaluate for chromosomal abnormalities and TORCH (toxoplasmosis, other agents, rubella, cytomegalovirus, herpes simplex) infections. To meet inclusion criteria, all fetuses had to have a thoracic abnormality, have no infection, have an absence of lethal genetic abnormalities and have a normal karyotype. In most cases, either the predominant cyst was aspirated and drained by a single puncture or a fetal thoracentesis was performed. The fetuses that had recurrence of dominant cysts or reaccumulation of pleural effusions were closely monitored with weekly ultrasound scans. All fetuses with type I CPAMs were given betamethasone in an effort to decrease the size of the CPAM. When fetal hydrops developed, emergent treatment with an in utero percutaneous thoracoamniotic shunt by the MFM-IR team was offered. The fetus was excluded from the study if an abnormal karyotype, other congenital malformation, or another underlying etiology for the development of nonimmune hydrops was found.

Maternal ages ranged from 22–33 years (mean, 27 y), and prior gestations ranged from 0–3 (mean, 1.4). All fetal thoracic abnormalities were discovered on ultrasound performed at gestational ages ranging from 20–24 weeks 2 days. Initial evaluations in the MFM clinic were performed between gestational ages of 23 weeks 4 days and 29 weeks 3 days. At the initial visit, ultrasound confirmed normal amniotic fluid indices (AFIs) in all patients. Three fetuses were found to have type I CPAMs, and two had chylothoraces. Type I CPAMs were characterized as macrocystic with a predominant large cyst. Four fetuses were 46XY, and one was 46XX. After initial cyst aspiration or thoracentesis, all of the cysts reaccumulated, or effusions recurred.

Percutaneous In Utero Thoracoamniotic Shunt Creation

Maternal anesthesia was obtained with an epidural, and fetal anesthesia was administered via intramuscular injection of vecuronium (0.2 mg/kg). Periprocedural antibiotic prophylaxis with cefazolin was administered. Under ultrasound guidance (LOGIQ E9; GE Medical Systems, Pewaukee, Wisconsin, or iU22; Philips, Andover, Massachusetts), an 18-gauge, 20-cm needle (INRAD, Inc, Kentwood, Michigan) was advanced via a percutaneous transabdominal, transuterine approach avoiding the placenta and umbilical cord into the fetus' thoracic abnormality (**Fig 2**). A 0.035-inch Rosen guide wire (Cook, Inc, Bloomington, Indiana) was coiled within the fetal thoracic abnormality (**Fig 3**). A 6-F, 13-cm peel-away sheath (Cook, Inc) was used as a buttress to stabilize the fetal chest wall. A 4.5-F, 6-cm double J ureteral stent (Gyrus ACMI; Southborough, Massachusetts) was modified by cutting off the trailing loop leaving a single loop at the leading edge for placement in the fetal thorax. This modification also allowed for a shorter shunt (6 cm) with known formed and unformed lengths. As the modified double J stent was advanced through the peel-away sheath, ultrasound