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Long-term outcomes after thoracoamniotic shunt for pleural effusions with secondary hydrops

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ABSTRACT

Background: Congenital pleural effusion is a rare condition with an incidence of approximately one per 15,000 pregnancies. The development of secondary hydrops is a poor prognostic indicator and such cases can be managed with a thoracoamniotic shunt (TAS). Our objective is to describe postnatal outcomes in survivors after TAS placement for congenital pleural effusions.

Materials and methods: A retrospective study of all cases with fetal pleural effusions treated between 2006 and 2016. Patients with dominant unilateral or bilateral pleural effusions complicated by secondary hydrops fetalis received TAS placement. The results are reported as median (range).

Results: A total of 29 patients with pleural effusion with secondary hydrops underwent TAS placement. The gestational age at the initial TAS placement was 27.6 (20.3-36.9) wk. Before delivery, hydrops resolved in 17 (58.6%) patients. The delivery gestational age was 35.7 (25.4-41.0) wk and the overall survival rate was 72.4%. Among the 21 survivors, 19 (90.5%) required admission to the neonatal intensive care unit for 15 (5-64) d. All 21 survivors had postnatal resolution of the pleural effusions. All 21 children were long-term survivors, with a median age of survivorship of 3 y 3 mo (9 mo-7 y 6 mo) at the time of last reported follow-up.

Conclusions: Thoracoamniotic shunting in fetuses with a dominant pleural effusion(s) and secondary hydrops resulted in a 72% survival rate. Nearly all survivors required admission to the neonatal intensive care unit. However, a majority did not have significant long-term morbidity.

Introduction

Congenital pleural effusions are a rare condition with an incidence of approximately one per 15,000 pregnancies.[1] Pleural effusions in the fetus can be unilateral or bilateral, and arise from various underlying etiologies. An isolated primary fetal pleural effusion is most commonly attributed to chylothorax.[2] Secondary etiologies may include pulmonary

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malformations, congenital heart disease, genetic syndromes, aneuploidy, fetal anemia, and infections.[2-4] The antenatal course is variable with spontaneous regression occurring in approximately 20% of primary fetal hydrothorax cases.[5] When congenital pleural effusions progressively enlarge and compromise fetal hemodynamics and cardiac function, secondary hydrops fetalis may develop.[6,7] The presence of hydrops is an important risk factor for poor prognosis.[5]

In an attempt to reduce the morbidity and mortality associated with pleural effusions complicated by secondary hydrops, these cases are often managed using a thoracoamniotic shunt (TAS).[2,4,5,8,9] However, owing to the rarity of this condition, there are only a few studies addressing the short- and long-term outcomes of fetuses after TAS placement.[8-15] The objective of this study was to describe postnatal outcomes in survivors after TAS placement for congenital pleural effusions with secondary hydrops.

Materials and methods

This was a retrospective study of all consecutive patients with a diagnosis of fetal pleural effusion treated at our center between 2006 and 2016. Each patient underwent a detailed ultrasound evaluation including fetal anatomy, Doppler waveforms (umbilical artery, umbilical vein, ductus venosus, middle cerebral artery), maximum vertical pocket of amniotic fluid, placental location and cervical length. The width of the pleural effusion was measured in the axial view at the level of the four-chamber view of the heart. Patients with a dominant unilateral or bilateral pleural effusion complicated by hydrops fetalis were offered TAS placement. The term "dominant" was reserved for fetuses with pleural effusions that were the first to develop or that were relatively more prominent than any remaining fetal effusions, and that were suspected to be the primary underlying cause of the cardiovascular compromise leading to the hydrops. Hydrops was defined as pleural effusion associated with one or more of the following: skin edema, pericardial effusion, ascites, or polyhydramnios (amniotic fluid maximum vertical pocket $\geq$ 8 cm).

Patients with structural anomalies noted on ultrasound were not excluded from TAS placement, but they were counseled that in such cases, the long-term prognosis may depend on an underlying genetic syndrome. A preoperative genetic amniocentesis was not required before TAS placement, but the amniotic fluid was sampled during shunt placement and sent for karyotype if this had not previously been performed. Noonan syndrome testing and whole genome single nucleotide polymorphism (SNP) microarray were not performed universally. Fetal genetic abnormality, such as trisomy 21, was not an exclusion criterion for TAS placement. A preoperative fetal thoracentesis was not routinely offered before TAS placement at our center.

TAS placement was performed under ultrasound-guidance using the Harrison shunt (Cook Medical). In brief, the 13-gauge trocar cannula system was inserted into the fetal pleural cavity. The trocar was then removed and, when technically feasible, an aliquot of the pleural fluid was aspirated using a 5-mL syringe. The pleural fluid was sent for culture, cell count (total nucleated cells, red blood cells, neutrophils, mononuclear cells), lactate dehydrogenase, protein, and glucose. The mononuclear cell count consisted of a total count of monocytes/histiocytes, lymphocytes, siderophages and erythrophages. The double pigtail Harrison shunt was then inserted into the fetal chest using the 5-French positioner. Ultrasound was used to confirm correct placement as well as efflux of pleural fluid into the amniotic cavity through the distal end of the shunt using color Doppler interrogation. If the initial shunt did not immediately begin draining the pleural fluid, or the shunt position was suboptimal, consideration was given to placement of a second TAS on the ipsilateral side. In cases of bilateral pleural effusions, the larger pleural effusion was initially targeted. Another shunt was placed on the contralateral side at the time of the initial surgery if there was still a large pleural effusion and/or mediastinal shift present after the initial TAS placement. If there was subsequent enlargement of the contralateral pleural effusion at a later time, the patient was readmitted for an additional TAS procedure.

Patients remained hospitalized for 24 h after surgery to monitor for postoperative complications and were discharged on postoperative day 1 after a repeat detailed ultrasound examination. Patients were managed for the remainder of the pregnancy by the referring perinatologist with weekly ultrasound examinations to assess for shunt location and resolution of hydrops. Patients with a dislodged or malfunctioning shunt were referred back for possible repeat TAS placement.

Data that were prospectively collected included maternal demographics, preoperative and postoperative ultrasound findings, surgical variables (gestational age [GA], number of shunts, pleural fluid analysis, complications, etc), and delivery outcomes. Long-term postnatal outcomes were obtained by the research team via acquisition of medical records and telephone inquiry of the patient. Patient characteristics and outcome data were analyzed univariately, and those patients who did and did not have neonatal survivors were compared with bivariate analysis. All analyses were performed using SAS statistical software (Version 9.2, SAS Institute Inc, Cary, NC). The results are reported as median (range). All study participants provided informed consent. This study was approved by the Health Sciences Institutional Review Board of the University of Southern California (Fetal Intervention Protocol: HS-16-00468), and complied with all patient protection criteria stipulated therein.

Results

A total of 29 patients (27 singleton and 2 twin gestations) with dominant pleural effusions with secondary hydrops were included: 21 (72.4%) bilateral and 8 (27.6%) unilateral (Table 1). The median preoperative pleural effusion width on the left and right were 2.1 (0.6-3.6) cm and 2.1 (0.4-5.0) cm, respectively. The GA at diagnosis was 27.4 (20.1-36.9) wk. Four (13.8%) patients underwent a thoracentesis by the referring perinatologist, and in all cases there was reaccumulation of the pleural effusions before initial TAS placement. The pleural effusions reaccumulated within 3-5 d in all patients.

Karyotype analysis from amniotic fluid was performed on all patients, yielding 28 (96.6%) euploid and 1 (3.4%) 47,XX,+21. The latter patient had a dichorionic twin gestation with a cell-