

Single Uterine Access for Bilateral Pleuroamniotic Shunting in Fetuses with Severe Hydrothorax by an Internal Rotational Maneuver: Feasibility and Outcomes between Successful and Failed Procedures

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Keywords

Hydrothorax · Hydrops · Pleural effusion · Pleuroamniotic shunt

Abstract

Objective: The objective of this study was to describe the feasibility of single percutaneous uterine access for bilateral pleuroamniotic shunting (PAS) in fetuses with severe hydrothorax by using an internal rotational maneuver and to compare perinatal outcomes between successful and failed procedures. **Methods:** A prospective cohort of 25 fetuses with isolated bilateral hydrothorax and hydrops were referred to our fetal surgery center in Queretaro, Mexico during an 8-year period. Bilateral PAS was first attempted through a percutaneous single uterine access by internal rotation of the fetus, which was achieved by using the blunt tip of the same cannula, and in case of a failed procedure, a second uterine port was used to place the second shunt. The perinatal outcomes between successful (single uterine port) and

failed (2 uterine ports) fetal procedures were compared. **Results:** Placing of bilateral shunts through a percutaneous single uterine access was feasible in 15/25 (60%) cases. Overall, median GA at delivery was 35.2 weeks with a survival rate of 64.0% (16/25). Three cases were excluded due to shunt dislodgement, leaving a final population of 22 fetuses; 13/22 (59.1%) and 9/22 (40.9%) managed using 1 and 2 uterine ports, respectively. The group with bilateral PAS placement through a successful single uterine port showed a significantly higher GA at birth (36.5 vs. 32.8 weeks, $p = 0.001$), lower surgical time (11.0 vs. 19.0 min, $p = 0.01$), longer interval between fetal intervention and delivery (5.7 vs. 2.7 weeks, $p = 0.01$), lower risk of preterm delivery (46.2 vs. 100%, $p < 0.01$), and lower rate of perinatal death (15.4 vs. 55.6%, $p < 0.05$) than the failed procedures requiring 2 uterine ports. **Conclusion:** In fetuses with severe bilateral hydrothorax and hydrops, bilateral pleuroamniotic shunting through a successful single percutaneous uterine access is feasible in up to 60% of cases and is associated with better perinatal outcomes.

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Introduction

Fetal hydrothorax is a congenital condition with an estimated incidence of 1 per 15,000 newborns [1] that can be primarily due to leakage of chyle from a lymphatic abnormal development [2] or secondarily due to different conditions, including genetic syndromes [3], fetal infections [4], and structural abnormalities [5, 6]. Prognosis of isolated primary hydrothorax without chromosomal abnormalities depends on different factors such as the gestational age (GA) of diagnosis, the existence of unilateral or bilateral pleural effusion, polyhydramnios as a consequence of fetal swallowing difficulties, and fetal hydrops which leads to a higher risk of intrauterine death. Fetuses with bilateral massive pleural effusions and hydrops are considered at the highest risk of perinatal death [1, 3]. For those cases, bilateral drainage of pleural effusions by bilateral pleuroamniotic shunt (PAS) placement has demonstrated to be an effective fetal therapy to reverse fetal hydrops and improve perinatal survival [7–10].

However, in the presence of bilateral massive pleural effusions, the literature does not offer homogeneous data about some technical aspects such as the number of required intrauterine accesses to place the bilateral pleuroamniotic shunts. Since preterm delivery of fetuses with pulmonary hypoplasia secondary to hydrothorax has been considered one of the main contributors to perinatal mortality [11, 12], it could be expected that a fewer number of intrauterine ports can lead to a lesser risk of preterm delivery, greater GA at birth, and thus higher sur-

vival probabilities. In keeping with this argument, placement of bilateral PAS through a single uterine port has been used as the first approach by several fetal surgery centers, which used a second uterine port to place the second shunt only when rotation fails [5, 8]. However, the feasibility of this approach, its technical details, and outcomes of the failed procedures are still unclear. Thus, the aim of this study was to describe the feasibility of single percutaneous uterine access for bilateral PAS in fetuses with severe bilateral hydrothorax and hydrops by using an internal rotational maneuver and to compare survival outcomes between successful and failed procedures.

Methods

During the study period between January 2012 and August 2020, a prospective cohort of consecutive fetuses with confirmed severe bilateral hydrothorax and hydrops were selected for fetal intervention with bilateral pleuroamniotic shunting in a single referral surgery center at Queretaro, Mexico. Detailed fetal morphological ultrasonography, advanced echocardiography, and fetal karyotype were performed in all cases.

All ultrasound examinations were performed by one of the 2 experienced examiners (R.C.M. or M.M.R.) using either a Voluson E8 Expert BT12.0 or Voluson E10 BT18.0 ultrasound machine equipped with a 6- to 2-MHz linear curved-array transducer. GA was determined by first or second trimester ultrasound examination. Inclusion criteria for PAS were singleton pregnancies with severe bilateral fetal hydrothorax, hydrops, and normal karyotype. Severe bilateral hydrothorax was defined as massive pleural effusion occupying >50% of both thoracic cavities, together with mediastinal shift and severe bilateral lung compression (Fig. 1). Hy-

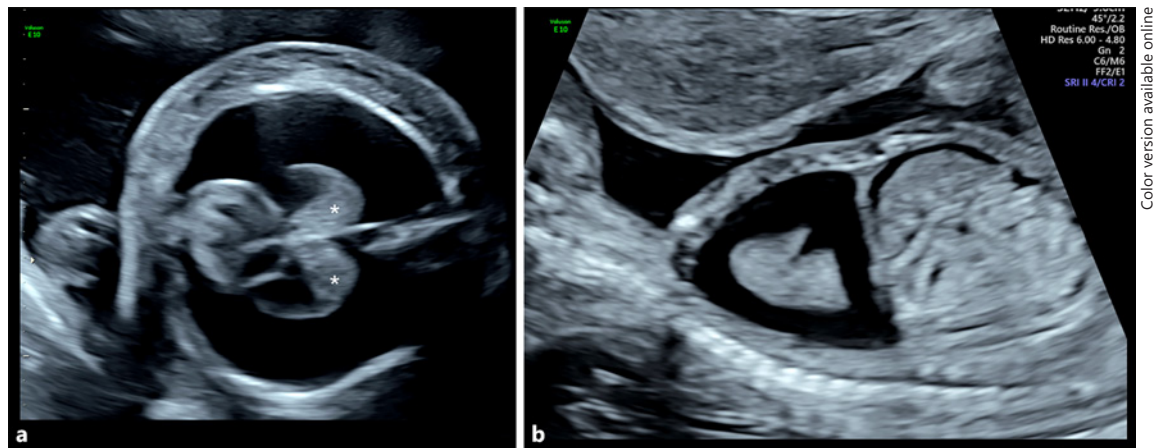


Fig. 1. Ultrasound picture of a 30-week fetus, with hydropic features (hydrothorax, ascites, and subcutaneous edema) in transversal (a) and sagittal (b) views of the thorax, demonstrating bilateral massive pleural effusion, severe bilateral lung compression (*), and mediastinal shift, considered candidate for bilateral pleuroamniotic shunting.

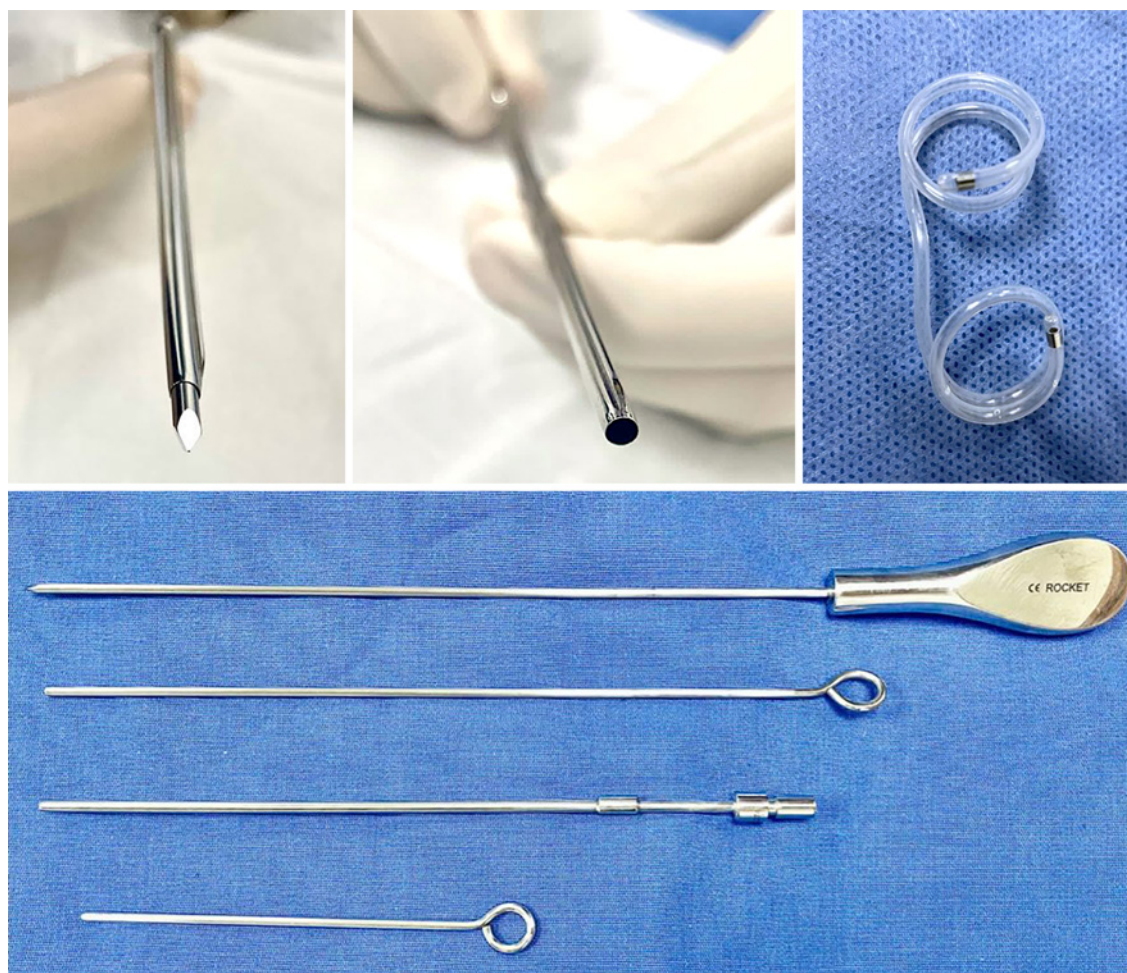
drops was defined as pleural effusion associated with either subcutaneous edema, ascites, or pericardial effusion [13]. Polyhydramnios was defined as an increased amniotic fluid volume with a deepest vertical pool ultrasound measurement above 8.0 cm [14]. The exclusion criteria were as follows: (a) preterm prelabor rupture of membranes (PPROM), (b) chromosomal abnormalities, (c) other structural anomalies, (d) placenta previa, and (e) short cervical length (<25 mm). Fetuses with hydrothorax secondary to lung lesions, congenital diaphragmatic hernia, or chromosomal abnormalities were also excluded as well as those with bilateral hydrothorax who required only unilateral PAS or those who required repetitive intrauterine intervention for persistent hydrothorax or catheter dislodgment.

Termination of pregnancy was not considered an option since it is not legal in our country after 12 weeks of gestation. The surgical protocol was approved by the hospital ethics committee (IRB 081/21-06-2017). Following counseling regarding the poor prognosis, the risk of intrauterine fetal demise associated with the presence of fetal hydrops, and the risks of the suggested fetal interven-

tion, the parents opted for bilateral PAS placement and signed an informed consent form. Patients were also informed about the potential risk of subsequent shunt blockage or dislodgment and the need to repeat fetal intervention.

Fetal Pleuroamniotic Shunting

Fetal intrauterine intervention was performed by a fetal medicine specialist (R.C.M.) with a formal training in fetal surgery. Before fetal intervention, the fetal position was evaluated by ultrasound and, if necessary, the fetus was moved by external manipulation to reach the optimal position, that is, a longitudinal position, with the fetal thoracic spine located against the uterine wall and in the contralateral side of the placenta. All fetal interventions were performed under maternal local anesthesia. For fetal anesthesia, fentanyl (15 mg/kg), vecuronium (0.2 mcg/kg), and atropine (0.2 mcg/kg) were injected into the fetal leg or arm under ultrasound guidance using a 22-gauge needle. Pleuroamniotic shunting was performed as previously described by Rodeck et al. [15]. Briefly, under ultrasound guidance, a 5F trocar was introduced percutane-



Color version available online

Fig. 2. Illustrative picture showing all the material used in pleuroamniotic shunts including the double pig-tail catheter, the 5F trocar inside the cannula, and the blunt tip of the cannula after trocar removal, which is used in the fetal rotational maneuver.

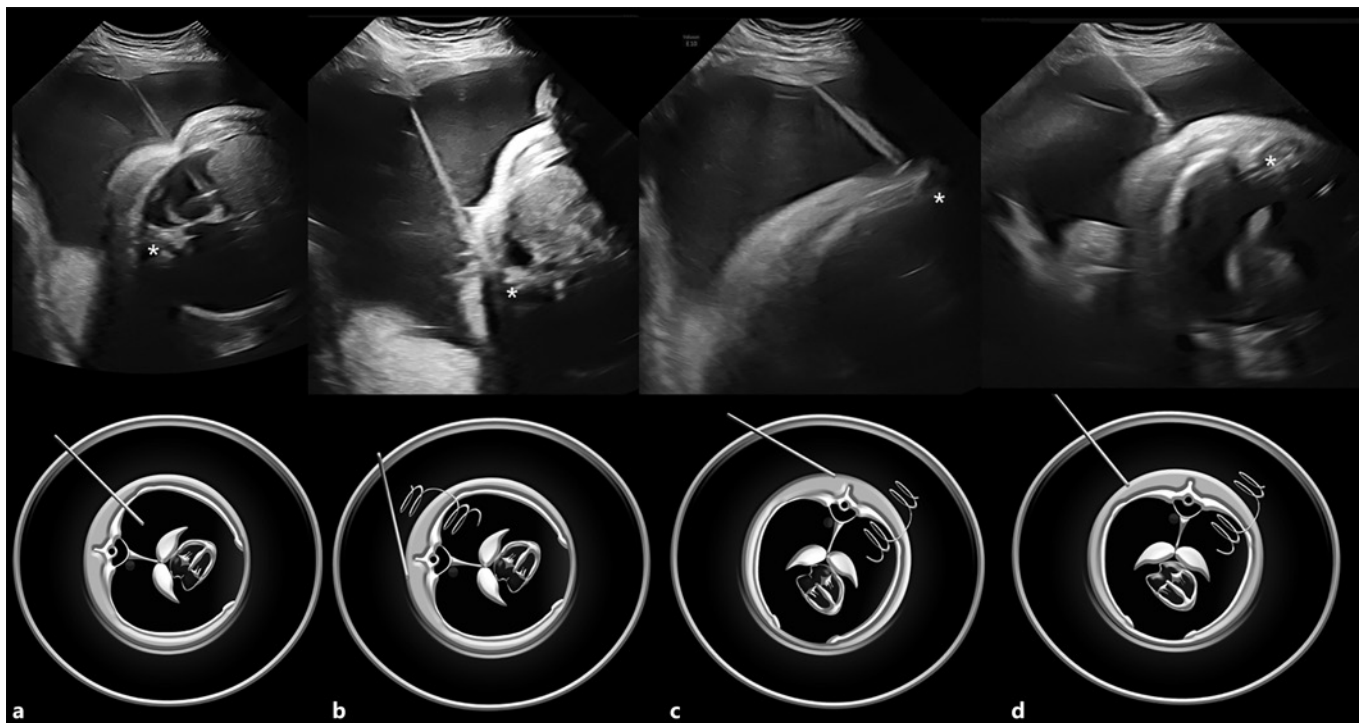


Fig. 3. Bilateral pleuroamniotic shunting by a single percutaneous uterine access. After placement of the first pleuroamniotic shunt (a), internal rotation (b, c) of the fetal back (*) was performed using the blunt tip of the cannula to approach the contralateral fetal hemithorax (d).

ously throughout the maternal abdomen into the amniotic cavity and advanced into the fetal thorax throughout the posterior intercostal space, and then a double-pigtail catheter (Rocket of London, London, UK) was placed in the fetal pleural space, releasing the distal portion into the amniotic cavity. The second pleuroamniotic shunt was placed in the contralateral hemithorax, employing the same percutaneous uterine port with internal rotation of the fetus achieved by using the blunt tip of the same cannula (Fig. 2), initiating the rotational movement in the contralateral side of the first catheter, making traction against the fetal back, and applying short sweeps until the second hemithorax was oriented in front of the sheet (Fig. 3). When the internal manipulation of the fetus was not feasible, that is, it took >3 attempts to rotate the fetal back (60 s per attempt) or the maneuver was lasting >3 min, we decided to place the second shunt by performing a second percutaneous uterine access and repeating the same technique used for the first shunt placement.

After fetal intervention, the patient was kept hospitalized for 24 h. Prophylactic tocolysis with indomethacin (100 mg every 12 h) or nifedipine (20 mg every 8 h) was administered during fetal surgery and up to 72 h after the procedure. Betamethasone (12 mg 2 doses 24 h apart) was administered for fetal lung maturation if the procedure took place between 28 and 36 weeks of GA. Location and correct placement of both catheters into the pleural spaces was confirmed by ultrasound during fetal follow-up, and, in case of detecting shunt blockage or dislodgment, a new shunt placement was performed. Cesarean delivery with clamping and withdrawal

of both pleuroamniotic catheters was planned after 38 weeks. Information regarding perinatal outcomes was collected in all cases including PPROM (defined as membrane rupture before labor that occurs before 37 weeks of gestation), intrauterine fetal demise, GA at delivery, interval between fetal intervention and delivery, neonatal death (defined as death within the first 28 days after birth), and perinatal death (defined as having either intrauterine fetal demise or neonatal death).

Statistical Analysis

All data were collected in an Access file for Windows 2010 (Microsoft Corp., Redmond, WA, USA). In order to determine the effect of the PAS technique on survival, the whole population was divided into 2 study groups. Group 1 was considered when a single uterine port was required for placement of both (left and right) shunts. Group 2 was considered when 1 intrauterine trocar insertion was required for each shunt placement, that is, 2 uterine ports. Survival outcomes and complications were compared between the 2 study groups. *T* test and χ^2 test were used to compare quantitative and qualitative data, respectively. Statistical calculations and descriptive analyses were performed using the Statistical Package for the Social Sciences (SPSS 25.0, SPSS Inc., Chicago, IL, USA) software. All tests were 2-tailed, and a probability value of <0.05 was considered statistically significant.

Table 1. Maternal and perinatal clinical characteristics of the studied population of fetuses with severe bilateral hydrothorax and hydrops treated with bilateral PAS either by a single or two-sided uterine access

Characteristic	Bilateral PAS by single uterine access, <i>n</i> = 13	Bilateral PAS by two-sided uterine access, <i>n</i> = 9	<i>p</i> value*
Maternal age, years	26.6 (6.3)	27.9 (7.9)	0.67
BMI, kg/m ²	26.3 (3.5)	26.9 (5.0)	0.75
Primiparity, %	23.1	55.6	0.12
Anterior placenta, %	46.2	55.6	0.67
Polyhydramnios, %	69.2	88.9	0.28
Cervical length, mm	32.5 (9.4)	28.5 (5.7)	0.27
GA at shunt, weeks	30.8 (2.2)	30.1 (3.1)	0.53
Shunt surgical time, min	11.0 (4.8)	18.9 (9.1)	0.01
Spent time for fetal manipulation, min	2.6 (0.5)	2.9 (0.2)	0.09
Amnioreduction, %	53.8	88.9	0.08
Amniodrainage, mL	353 (393)	351 (352)	0.99
PPROM, %	15.4	22.2	0.68
GA at PPROM, weeks	34.8 (0.5)	34.6 (0.6)	0.74
Interval between shunt and birth, weeks	5.7 (2.9)	2.7 (2.3)	0.01
GA at delivery, weeks	36.5 (1.6)	32.8 (2.9)	<0.01
Cesarean delivery, %	92.3	66.7	0.13
Preterm delivery <37 weeks, %	46.2	100.0	<0.01
Preterm delivery <34 weeks, %	7.7	55.6	0.01
Birth weight, g	3,130 (784)	2,207 (810)	0.02
Neonatal death, %	15.4 (2/13)	50.0 (4/8)	0.08
Perinatal death, %	15.4 (2/13)	55.6 (5/9)	0.04

GA, gestational age; PPROM, preterm premature rupture of membranes; PAS, pleuroamniotic shunting.
* Student's *t* test for independent samples or Pearson- χ^2 test.

Results

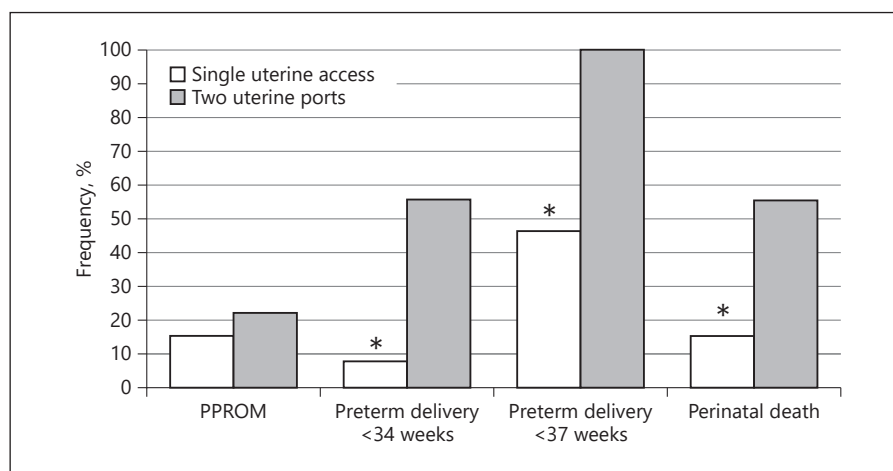
A total of 96 consecutive fetuses with hydrothorax were referred to our center during the study period; of these, 25 with isolated bilateral hydrothorax, hydrops, and normal karyotype were selected for bilateral PAS. Bilateral PAS was successfully performed in all cases at a median GA of 30.7 (25.7–34.0) weeks. The median surgical time was 13.0 (range, 5.0–33.0) minutes. Bilateral PAS was successfully performed through a single uterine access in 15/25 (60%) of the cases. No surgical complications such as chorioamnionitis, placental abruption, or uterine bleeding were observed. Preterm rupture of the membranes occurred in 6/25 (24.0%) cases at a median GA of 34.7 (34.1–35.1) weeks. The median GA at delivery was 35.2 (range, 25.9–38.6) weeks. There were 2 intra-uterine fetal demises (8.0%) and 7 neonatal deaths (28.0%). Thus, overall survival rate was 64.0% (16/25). In 3 cases, PAS had to be repeated due to shunt dislodgment. Therefore, those cases were excluded for final analysis, leaving a final population of 22 fetuses; on which, bilat-

eral pleuroamniotic catheters were placed by a single percutaneous uterine access in 13/22 (59.1%) fetuses and through 2 uterine ports in 9/22 (40.9%) fetuses.

Table 1 shows the baseline maternal and fetal characteristics between the study groups. No differences were observed in GA at fetal intervention, time spent for fetal manipulation, rate of polyhydramnios, PPROM, and mode of delivery. The group with severe hydrothorax and bilateral shunting placed through a single uterine port showed a significantly lower surgical time (11.0 vs. 18.9 min, $p = 0.01$), longer interval between fetal intervention and delivery (5.7 vs. 2.7 weeks, $p = 0.01$), and higher GA at birth (36.5 vs. 32.8 weeks, $p = 0.001$) than those placed through 2 uterine ports.

Figure 4 shows the perinatal outcomes of fetuses with severe bilateral hydrothorax and hydrops treated with bilateral PAS by using either a single or 2 uterine ports. In comparison to the group with 2 uterine ports, those with a single uterine access showed a significantly lower incidence of delivery within 1 week after PAS (0 vs. 33.3% [3/9], respectively, $p = 0.02$), lower risk of preterm deliv-

Fig. 4. Proportion of adverse perinatal outcomes in fetuses with severe bilateral hydrothorax and hydrops treated with two-sided PAS by using either a single or 2 percutaneous uterine ports. (*) statistically significant ($p = <0.05$). PAS, pleuroamniotic shunting.



ery at <37 (46.2% [6/13] vs. 100% [9/9], $p < 0.01$) and 34 (7.7% [1/13] vs. 55.6% [5/9], $p = 0.01$) weeks, lower rate of neonatal death (15.4% [2/13] vs. 50.0% [4/8], $p = 0.08$), and significantly lower rate of perinatal death (15.4% [2/13] vs. 55.6% [5/9], $p < 0.05$).

Discussion

This study reports the technique, feasibility, and perinatal outcomes of single percutaneous uterine access for bilateral pleuroamniotic shunting in fetuses with severe bilateral hydrothorax and hydrops. We found that placement of bilateral PAS by a single uterine port was feasible in up to 60% of the cases, and, in comparison to shunts placed through 2 uterine entrances, single uterine access for bilateral PAS was associated with better perinatal outcomes.

This is not the first case series that included fetuses with severe bilateral hydrothorax treated with PAS employing a single uterine access. This technique was first published by Nicolaides et al. [5] in 1990. In such study, the authors included 25 fetuses with bilateral hydrothorax treated with PAS, but the number of cases with bilateral shunting and those placed by a single uterine access were not described. In a more recent study published by Yinon et al. [8], the proportion of cases with bilateral PAS through a single percutaneous uterine port was higher than that observed in our study. The authors inserted bilateral shunts in 41/54 (75.9%) fetuses, and notably 5 (12.2%) of them were placed without the need of fetal rotation. Although this observation was not clarified by the authors, it could be explained if some shunts were placed through the anterior wall of the fetal thorax as it has been previously reported by the same authors in their original description of the technique [16]. In our study

however, all PAS were placed through the posterior wall of the fetal thorax in an attempt to decrease the risk of shunt dislodgment. Another potential explanation to our lower successful rate of bilateral PAS by a single uterine port could be the different GA at fetal intervention between both groups. While in the former study the median GA at shunt placement was 27.6 weeks, it was 30.5 weeks in our study; thus, it could be argued that placement of bilateral shunts by a single uterine access might be more challenging at advanced GAs.

No previous studies have compared the perinatal outcomes of cases treated with bilateral PAS through a single or double uterine access. Our study suggests that bilateral PAS placement by a successful single uterine port might be less invasive since it was associated with both higher interval between fetal intervention and delivery, and significantly higher GA at birth than failed procedures requiring 2 uterine ports. A pathophysiological explanation of our findings might be related to the fewer intrauterine incisions and less intrauterine surgical time in the group with a single uterine access. Even though decompression of the mediastinum with bilateral PAS alleviates the cardiac repercussions caused by severe hydrothorax regardless of the number of uterine ports, a lower incidence of preterm delivery and better perinatal survival were observed in those managed with a single uterine access.

Strong evidence has demonstrated that similar to other fetal lung anomalies, avoiding preterm delivery in fetuses with massive pleural effusions is crucial to improve neonatal survival and decrease neonatal respiratory morbidity. In agreement with this argument, Dorsi et al. [12] reported the relationship between higher interval between PAS and birth with better chances of reversal of hydrops and neonatal survival. Similarly, Bianchi et al.

[17] showed that delivery after 35 weeks is associated with less need of mechanical ventilation among fetuses that were offered PAS for severe hydrothorax and survived; therefore, it could be expected that the reduction of the uterine incisions might be clinically useful in decreasing the risk of preterm delivery and improving neonatal survival. In this contention, Derderian et al. [18] demonstrated that the number of intrauterine procedures affect neonatal outcomes. The authors included 21 fetuses with hydrothorax treated with PAS and showed higher survival rates in those with 1 or 2 fetal procedures than those with 3 or more intrauterine interventions (90 vs. 20%, $p < 0.01$, respectively). Similarly, the rate of preterm delivery was higher in the group with 3 or more fetal interventions (100%) in comparison to those with 2 procedures (50%) and those with a single fetal procedure (38%).

The main strength of this study was the inclusion of a well-selected population of fetuses with isolated severe bilateral hydrothorax and hydrops, requiring bilateral shunting and the exclusion of pregnancies with short cervical length that may bias the results by increasing the risk of preterm delivery. Our study does have limitations; first of all, the small sample size did not allow us to compare the rate of shunt dislodgment between the study groups. Second, we recognize that not all fetuses with bilateral hydrothorax require placement of bilateral shunts since a proportion of cases might be managed with a single pleuroamniotic catheter placed in the dominant side of the effusion. Those cases were not considered in the current study because they represent a less severe group, similar to those with unilateral hydrothorax. We also recognize that bilateral shunting utilizing a single uterine access with internal rotation of the fetal back to approach the contralateral hemithorax might be very challenging since even in experienced hands up to 40% of the cases required a second uterine access; therefore, a formal training on the aforementioned intrauterine maneuver would be needed in order that fetal surgeons acquire the necessary skills to perform such intervention within a short intrauterine time. Finally, we recognize that fetal manipulation for unsuccessful fetal rotation could be a potential factor that may explain the worst results observed in the group with 2 uterine accesses. However, while all cases managed with 2 ports had additional time because of unsuccessful fetal manipulation, up to 80% of the shunts placed through 1 uterine port also required >1 attempt to allow fetal rotation and consequently required additional surgical time. Thus, a randomized control trial would add more information to confirm our findings, where instead of spending time for unsuccessful fetal rotation, a group would be predetermined to have either 1

or 2 uterine accesses, this way ensuring more homogeneous populations for comparison.

In conclusion, bilateral pleuroamniotic shunting through a single percutaneous uterine access is feasible in up to 60% of the cases and is associated with better survival outcomes. Based on our results, we propose that among fetuses with severe bilateral hydrothorax and hydrops candidates for bilateral PAS, a single percutaneous uterine access could be attempted as the first approach, and routine bilateral intrauterine access avoided, as a way to decrease the risk of preterm delivery and perinatal death. However, further studies with higher sample size and more homogeneous populations are required to confirm our results before incorporating this recommendation into clinical practice.

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Statement of Ethics

Our research complies with the guidelines for human studies and was conducted ethically in accordance with the World Medical Association Declaration of Helsinki. All subjects gave their written informed consent, and the study protocol was approved by the Hospital Ethics Committee (Hospital de Especialidades del Niño y la Mujer Dr. Felipe Núñez Lara, Querétaro, México IRB 081/21062017).

Conflict of Interest Statement

The authors have no conflicts of interests to declare.

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Author Contributions

R.C.M., M.M.R., A.G.V., H.L.B., and J.L.G. participated in all the fetal interventions. R.C.M. and C.S. made substantial contributions to design the study protocol. E.C.R., A.G.V., and I.J.M. carried out acquisition of data. R.C.M. and E.C.G. performed the analysis and interpretation of data. M.M.R. and R.V.G. drafted the manuscript. All authors listed revised the article for important intellectual content and gave final approval of the final version for publication.

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