SCHD
Structural & Congenital Heart Disease

Tech Science Press

http://doi.org/10.32604/schd.2026.079259

CASE REPORT

Successful Ductus Arteriosus Stenting Using a Veno-Arterial Loop for Severe Pulmonary Hypertension in an Infant: A Case Report

Taro Kono, Naofumi F. Sumitomo*, Atsushi Maruyama and Takayuki Oyanagi

Department of Pediatrics, Keio University School of Medicine, Tokyo, Japan
*Corresponding Author: Naofumi F. Sumitomo. Email: naosumi1983@keio.jp

Received: 18 January 2026; Accepted: 06 July 2026; Published: 31 July 2026

ABSTRACT: Background: A reverse Potts shunt is performed as a palliative intervention for severe pulmonary hypertension (PH) to decompress the right ventricle. Several reports have described transcatheter stenting of the patent ductus arteriosus (PDA) to create an endogenous Potts shunt in infants with severe PH, typically via the femoral vein approach. **Case Description:** We present the case of a 7-month-old, 7.5 kg female infant diagnosed with severe PH dependent on PDA circulation, who was treated with PDA stenting via the right internal jugular vein (RIJV) approach due to inferior vena cava occlusion. A veno-arterial wire loop was created from the femoral artery to the RIJV to stabilize the wire and stent delivery. A 6 × 14 mm stent was successfully inserted into the PDA without complications. **Conclusion:** The RIJV approach using the veno-arterial loop technique is a viable and effective treatment for severe PH in infants with limited vascular access.

KEYWORDS: Ductal stenting; Potts shunt; infant; ductus arteriosus; pulmonary hypertension

1 Introduction

Pulmonary arterial hypertension (PAH) is a fatal disease in which increased pulmonary vascular resistance restricts pulmonary blood flow, leading to right-sided heart failure. Severe pulmonary hypertension (PH) in infants often progresses to a ductus-dependent state. In cases refractory to medical therapy, lung transplantation may be required [1,2].

Recently, the reverse Potts shunt, which creates a connection between the aorta and pulmonary artery (PA), has been reported as a palliative intervention to bridge patients with lung transplantation [3]. This shunt decompresses the right ventricle (RV) and maintains cardiac output, even in the setting of severely elevated PA pressure. Transcatheter patent ductus arteriosus (PDA) stenting has recently been reported as an endogenous reverse Potts shunt procedure in infants with severe PH [4–6]. In such cases, an approach through the femoral artery and vein is usually attempted [7]. However, if the inferior vena cava (IVC) is occluded, an alternative approach should be considered. Herein, we report a case of severe PH with IVC obstruction in which transcatheter PDA stenting was successfully performed by creating a veno-arterial (VA) loop between the internal jugular vein and the femoral artery.

2 Case Presentation

A female neonate with intrauterine growth restriction (IUGR) and a birth weight of 1721 g (1.3 percentile, −2.23 SD), length of 42.3 cm (4.2 percentile, −1.73 SD), and head circumference of 29.1 cm (1.9 percentile,

−2.08 SD) was delivered via caesarean section at 36 weeks and 2 days of gestation, consistent with symmetrical IUGR with multiple congenital anomalies. She required mechanical ventilation soon after birth because of respiratory failure. Transthoracic echocardiography showed main pulmonary artery (MPA) dilation with a diameter of 12.0 mm (Z-score = +3.5) and bilateral hypoplastic PAs: the left PA measured 2.2 mm (Z-score = −3.1) and the right PA 2.9 mm (Z-score = −1.8). Other findings included an atrial septal defect, mild coarctation of the aorta, and a PDA with bidirectional laminar flow. On day 3, transthoracic echocardiography revealed an increased tricuspid regurgitation pressure gradient (TRPG) of 80 mmHg with right-to-left shunting across the narrowed ductus arteriosus. Thus, the patient was diagnosed with ductus-dependent severe PH. Echocardiography showed no evidence of increased pulmonary blood flow. Chest computed tomography (CT) revealed hypoplastic lungs and bilateral PAs, and whole-exome sequencing revealed a deletion in 14q32, which is associated with alveolar hypoplasia. Therefore, the patient was diagnosed with congenital severe PH secondary to bilateral hypoplastic PAs associated with alveolar hypoplasia related to the 14q32 deletion. After sequential administration of lipo-prostaglandin E1 (20 ng/kg/min) and inhaled nitric oxide, the ductus arteriosus was recanalized, and the TRPG decreased to 50 mmHg. Her general condition was stabilized by day 14, and nitric oxide was switched to sildenafil. The PDA was opened to a maximum diameter of 4.6 mm with continuous prostaglandin E1 administration. In addition, she had craniosynostosis, a small jaw, and tracheomalacia and underwent tracheostomy at 3 months and cranioplasty at 4 months. Subsequently, she required continuous ventilator support due to weak spontaneous breathing.

At 5 months, an attempt was made to wean off prostaglandin-E1. Meanwhile, the TRPG increased to 120 mmHg with narrowing of the ductus arteriosus, indicating suprasystemic PH. Prostaglandin E1 tapering was halted, and transcatheter PDA stenting was planned to discontinue prostaglandin E1 at 7 months of age, when the patient weighed 7.5 kg.

Preoperative contrast-enhanced CT revealed IVC occlusion and cardiac dextroversion (Fig. 1a–c). Given that a femoral vein catheter approach was not feasible, a 5-French, 5-cm sheath was inserted through the right internal jugular vein (RIJV).

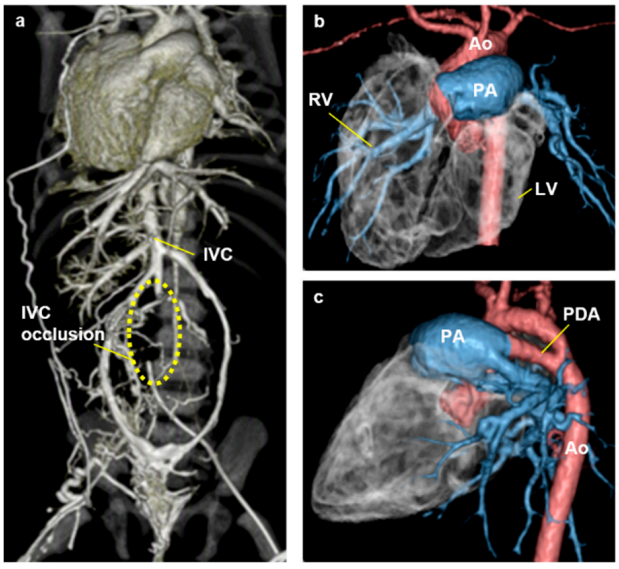

Figure 1: Contrast-enhanced computed tomography. (**a**) Dextroversion of the heart and inferior vena cava occlusion. (**b**) Frontal view shows right ventricular enlargement. (**c**) Lateral view shows patent ductus arteriosus, Krichenco type C. Ao, aorta; IVC, inferior vena cava; LV, left ventricle; RV, right ventricle; PA, pulmonary artery; PDA, patent ductus arteriosus.

Next, a 3.3-French sheath was inserted into the left femoral artery (LFA) to create a VA loop. The PDA had a minimum diameter of 4.1 mm at a slight ridge on the PA side and a length of 11 mm (Fig. 2a,b). A 0.014-inch guidewire was inserted into the MPA from the LFA sheath and captured from the RIJV sheath using a snare catheter (Fig. 2c,d). The wire was pulled out through the RIJV sheath, creating a VA loop with the following sequence: LFA→PDA→MPA→right ventricle→right atrium→RIJV. A 5-French multi-purpose guiding sheath (45 cm) was advanced from the RIJV through the PDA into the descending aorta, and an Express^{TM} Vascular SD stent measuring 6–14 mm (Boston Scientific, MA, USA) was deployed into the ductus arteriosus (Fig. 2e,f). Severe PAH was confirmed with a PA pressure of 79/41 mmHg (mean, 59 mmHg) and an ascending aorta pressure of 91/40 mmHg (mean, 61 mmHg). PA angiography revealed dilation of the MPA and narrowing of the peripheral PA. During catheterization, hypotension was managed with epinephrine and fluid infusion. Blood transfusion was administered for anemia related to the procedure and to maintain adequate circulating blood volume. Postoperatively, prostaglandin E1 was successfully discontinued. One and a half years after the procedure, no stent migration, narrowing of the ductus arteriosus, or other complications were observed. The cost of treatments during hospitalization in this case was covered under the Japanese universal health insurance system and local government medical expense subsidy programs for children.

This study was conducted in accordance with the Declaration of Helsinki. Written informed consent for the publication of this case report and accompanying images was obtained from the patient's legal guardian. Ethical approval was not required in accordance with the institutional policy for single case reports.

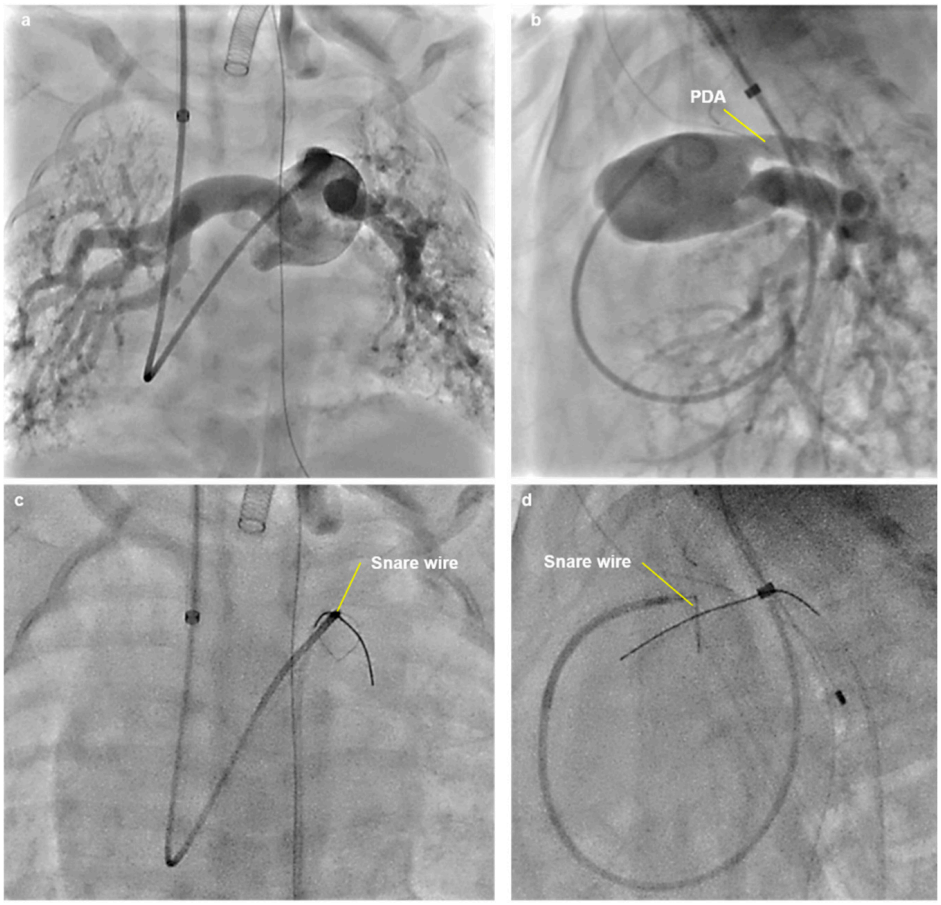

Figure 2: *Cont.*

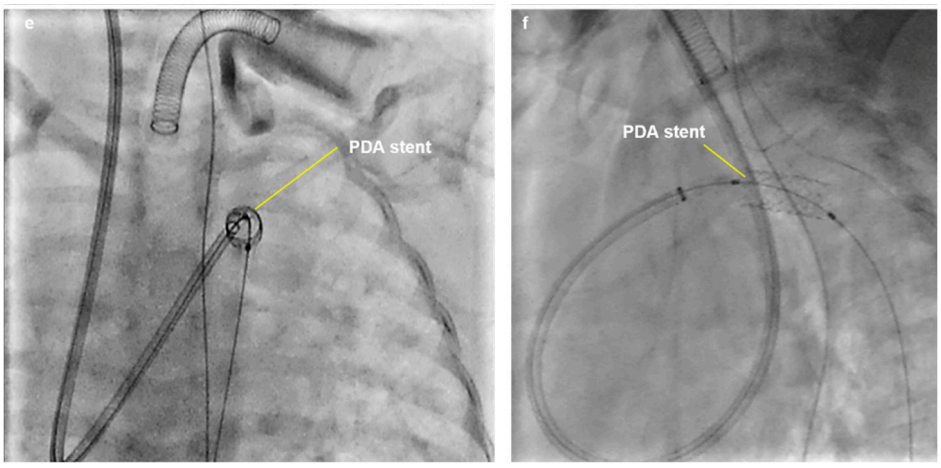

Figure 2: Angiography. (**a**) Pulmonary artery angiography: frontal view shows dilation of the main pulmonary artery and narrowing of the peripheral pulmonary artery. (**b**) Lateral view shows PDA with the narrowest point of 4.1 mm at the pulmonary artery ridge and a length of 11 mm. (**c**,**d**) Snare catheter capturing the guidewire tip. (**e**,**f**) Express SD stent deployed in the ductus arteriosus. PDA, patent ductus arteriosus.

3 Discussion

To our knowledge, this is the first case of PDA stenting to create an endogenous reverse Potts shunt via the RIJV using a VA loop technique in an infant with IVC obstruction. In cases of obstructed IVC, femoral venous access is challenging, and alternative access routes have been described [8]. However, a simple RIJV approach without a VA loop presents two major technical challenges: first, the guiding wire may lack stability due to a sharp hairpin bend at the RV; and second, the delivery sheath can compress the tricuspid valve, leading to hemodynamic instability compared with the usual femoral vein approach.

The instability of the delivery system increases the risk of sheath delivery difficulty, stent malposition, or procedural failure. Therefore, the development of a robust delivery system is necessary. As a method to stabilize guiding-sheath delivery, a VA loop from the femoral vein to the femoral artery has been reported in adult transcatheter valve implantation for mitral valve stenosis [9,10] and in pediatric valvuloplasty for aortic valve stenosis [11]. However, there have been no prior reports on the use of similar techniques for stent placement in the ductus arteriosus. A VA loop was adopted to overcome the instability of the RIJV approach. In our case, constructing an RIJV-femoral artery wire loop ensured delivery of both the long sheath and stent without major complications.

Hemodynamic compromise due to tricuspid valve interference remains a concern, even after creating a VA loop. When a long sheath crosses the tricuspid valve, circulatory failure can occur owing to increased tricuspid valve regurgitation. This risk has been documented not only in adult pulmonary valve replacement procedures [12], but also in infants undergoing transvenous interventions such as right ventricular outflow tract stenting [13]. In infant procedures, careful monitoring for early signs of circulatory deterioration is essential, and preparation for fluid infusion, blood transfusion, antiarrhythmic agents, and catecholamines is required to allow immediate intervention. In the RIJV approach, the tricuspid valve is positioned at the inflection point of the delivery system's hairpin curve, thereby further increasing the risk. In the present case, this risk was managed with epinephrine, fluid infusion, and blood transfusion.

Another possible approach in our case could have been the transarterial approach by placing a large, long sheath in an artery. Carotid, axillary, and femoral artery approaches have been reported as feasible in infants with ductal-dependent pulmonary blood flow, particularly in cases of ductal spasms or very tortuous

PDAs. In contrast, procedural complication rates of 18.2%–20.4% have been reported, including bleeding, thrombosis, aortic occlusion, and aortic dissection [14]. Therefore, when performing ductus arteriosus stent placement, a transvenous approach should be prioritized whenever possible [14]. In our case, the PDA was straight and wide, allowing selection of the RIJV approach with placement of only a small, short sheath in the femoral artery. No postoperative vascular complications were observed.

It is also important to consider the underlying causes of IVC obstruction. Abnormal cardiac position, including isomerism or dextroversion, is associated with systemic venous anomalies such as IVC interruption or stenosis [15]. In addition, prolonged central venous catheterization in low-birthweight neonates and infants with cardiac or other comorbidities can lead to thrombotic IVC occlusion [16,17]. Although no direct causal link has been established between the patient's 14q32 microdeletion and IVC obstruction, the combination of a possible congenital venous anomaly and catheter-related thrombosis likely contributed to IVC obstruction. These overlapping risks highlight the need for alternative and flexible access strategies. In this case, the selected strategy, an RIJV approach with a VA loop, effectively addressed anatomical challenges and enabled successful intervention without vascular complications.

Despite the successful outcome, several limitations should be acknowledged. First, as a single case report, the generalizability of this RIJV–femoral artery VA loop technique to other infants with severe PAH and IVC obstruction is inherently limited. Second, while the PDA stent remained patent at 1.5 years of follow-up, we relied primarily on echocardiography for surveillance; we did not perform routine follow-up cardiac catheterization or CT angiography to objectively quantify the long-term shunt fraction or accurately assess the residual right ventricular pressure burden. Third, although the VA loop stabilized the delivery system, this approach still requires femoral arterial access, which carries a non-negligible risk of vascular complications (e.g., thrombosis or limb ischemia) in small infants, despite the use of a small sheath. Lastly, the exact etiology of the IVC occlusion in this patient—whether congenital (associated with dextroversion) or acquired (secondary to prior catheterization)—remains speculative, as we lacked histological or detailed sequential imaging evidence to confirm the primary cause.

4 Conclusion

The RIJV approach with a VA loop technique is a feasible and effective alternative for PDA stenting in infants with severe PH when conventional femoral venous access is not possible because of IVC obstruction. This technique successfully stabilized the wire and stent delivery system, enabling precise stent placement without major vascular complications. The VA loop overcame the technical challenges of the RIJV approach, including wire instability and potential tricuspid valve interference. This case demonstrates that with careful hemodynamic monitoring and preparation for circulatory support, the RIJV-femoral artery VA loop technique can be successfully employed in small infants with complex vascular anatomy, including those with IUGR and multiple congenital anomalies.

Acknowledgement: The authors thank Drs. Masanori Inoue and Mariko Hida for their dedicated care of the patient.

Funding Statement: This research was funded by the JSPS KAKENHI grant No. JP 25K19211.

Author Contributions: The authors confirm their contributions to the paper as follows: Conceptualization, Naofumi F. Sumitomo; original draft preparation, Taro Kono; writing, review, and editing, Naofumi F. Sumitomo, Atsushi Maruyama and Takayuki Oyanagi; supervision, Naofumi F. Sumitomo. All authors reviewed and approved the final version of the manuscript.

Availability of Data and Materials: The data that support the findings of this study are available from the Corresponding Author, Naofumi F. Sumitomo, upon reasonable request.

Ethics Approval: This study was conducted in accordance with the Declaration of Helsinki. Written informed consent for the publication of this case report and accompanying images was obtained from the patient's legal guardian. Ethical approval was not required in accordance with the institutional policy for single case reports.

Conflicts of Interest: The authors declare no conflicts of interest.

Abbreviations

PH	Pulmonary Hypertension
PDA	Patent Ductus Arteriosus
MPA	Main Pulmonary Artery
RV	Right Ventricle
TRPG	Tricuspid Regurgitation Pressure Gradient
RIJV	Right Internal Jugular Vein
IVC	Inferior Vena Cava
LFA	Left Femoral Artery
VA	Veno-Arterial
Ao	Aorta
LV	Left Ventricle
PA	Pulmonary Artery
RVOT	Right Ventricular Outflow Tract

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