

Ductal flow reduction with covered coronary stents in neonates with pulmonary overflow after ductal stenting



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ABSTRACT

Ductal stenting (DS) is currently considered a reliable and safe alternative to surgery for maintaining duct patency in patients with duct-dependent pulmonary blood flow. However the presence of pulmonary overflow has been shown to complicate short-term progress in some patients. Although in most instances this complication is managed conservatively, serious events have been reported. We present two cases of pulmonary atresia and intact ventricular septum with pulmonary overflow after ductal stenting, which were successfully treated by implantation of multiple covered and bare metal coronary stents intra-stent.

1. Introduction

Surgical palliation, mainly in the form of systemic-to-pulmonary shunts, has for many years been the technique of choice to treat newborns and infants with duct-dependent pulmonary circulation. However, surgical palliation has been associated with high morbidity and mortality rates [1]. Ductal stenting (DS) was first used in the early 1990s [2,3] and is currently considered a reliable and safe alternative to surgery for maintaining duct patency in patients with duct-dependent pulmonary blood flow [4–7]. The DS success rate nears 100% and is associated with low complication rates when performed by experienced centers [4,8,9]. Pulmonary overflow is a rare but challenging complication after DS [4,7,9–11]. We discuss herein two cases of pulmonary atresia and intact ventricular septum with pulmonary overflow after ductal stenting, which were successfully managed by implantation of multiple covered and bare metal coronary stents intra-stent.

2. Case 1

Newborn male (weight 4.350 kg) with pulmonary atresia intact ventricular septum (PAIVS). Prostaglandin (PGE1) infusion was started at birth and at 48 h of life, the patient was taken to the catheterization laboratory for perforation of the pulmonary valve. The right ventriculography showed a tripartite right ventricle (RV) with well-developed pulmonary infundibulum and a pinhole patency of the pulmonary valve. No coronary fistulae were found. A balloon dilatation of the pulmonary valve was performed. Unfortunately, weaning from PGE1

was not possible. Hence, at day 23 of life (weight 4.440 kg), the decision was made to perform a DS. The initial aortic angiogram showed a straight arterial duct with a diameter of 5.6 mm at the aortic end, 3.5 mm at the pulmonary end and a length of 14 mm. The stenting procedure was performed via the femoral artery. Two coronary stents 4.5 mm × 15 mm and 4.5 mm × 18 mm (Driver® Sprint, Medtronic, Galway, Ireland) expanded to a 4.2 mm diameter according to the stent ATM chart were implanted. Soon after arriving to the PICU, the patient developed marked signs of pulmonary overflow requiring mechanical ventilation and inotropic support. After two days of conservative management, the patient was returned to the catheterization laboratory for a ductal flow reduction procedure. The initial angiography showed an internal diameter of the ductal stent of 4.2 mm. Through a 4-Fr sheath, via the femoral artery, a 4 × 12 mm coronary stent (Integrity®, Medtronic, Galway, Ireland) was inserted within the ductal stent. Subsequently, another coronary stent (Integrity® 2.5 mm × 8 mm) was mounted on a covered stent (3.5 mm × 18 mm) (Aneugraft® Dx, ITGI Medical, Or Akiva, Israel) with the aim of creating a “Diabolo stent” (Fig. 1A). Balloon inflation showed complete dilatation of both stents without the intended “diabolo” shape. Since this did not achieve the targeted O₂ saturation, it was decided to implant an additional Integrity® 3 mm × 9 mm stent (Fig. 1B and C). The duct diameter decreased from 4.2 mm to 3 mm. Arterial O₂ saturation and pO₂ decreased from 93% and 57 mm Hg to 79% and 41 mm Hg respectively after the procedure. Pulmonary overflow subsequently resolved and the patient was discharged four days later with a clinically stable condition.

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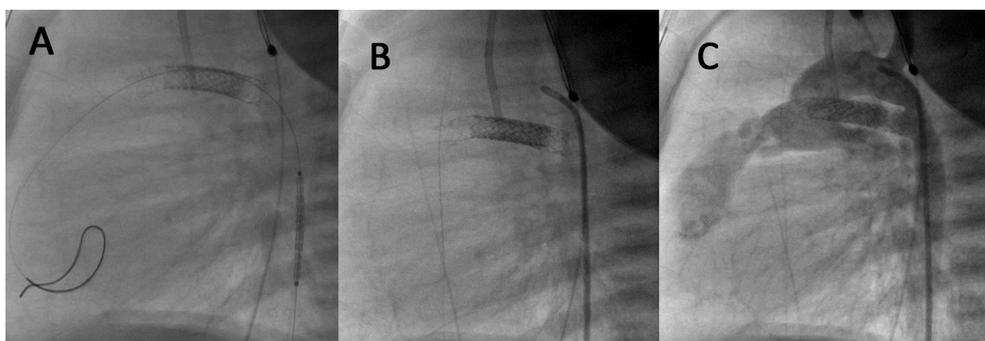


Fig. 1. A: Lateral fluoroscopic image showing the previously implanted ductal stents (Driver® Sprint 4.5 mm × 15 mm and 4.5 mm × 18 mm) traversed by a coronary 0.014" guidewire and the "Diabolo stent" assembly (Integrity® 2.5 mm × 8 mm mounted on a Aneugraft® Dx covered stent 3.5 mm × 18 mm) in the descending aorta. B: Lateral fluoroscopic image showing a reduction in ductal diameter as a result of all the implanted stents (two Driver® Sprint, two Integrity® and one Aneugraft® Dx). C: Final lateral aortic angiogram after the ductal flow reduction procedure.

3. Case 2

Newborn female (weight 3.300 kg) with diagnosis of PAIVS. Echocardiography showed a significantly hypoplastic RV and tricuspid valve with a small coronary fistula. PGE1 infusion was initiated and a DS was performed at 8 days of age. The initial aortic angiogram showed a tortuous duct arising from the aortic isthmus, with a diameter of 5.3 mm at the aortic end, 4.4 mm at the pulmonary end and a length of 15 mm. A 4.5 mm × 18 mm coronary stent (Driver® Sprint) expanded to its nominal diameter was successfully implanted in the arterial duct via the femoral artery. Soon after the procedure, the patient showed signs of pulmonary overflow and low cardiac output. She developed necrotizing enterocolitis as well as electrocardiographic changes and troponin elevation suggesting myocardial ischemia. A ductal flow reduction procedure was therefore performed. In the initial aortic angiography, the internal duct diameter was 4.4 mm. Four coronary stents of 4.5 × 15 mm (Driver® Sprint), 4 × 12 mm (Integrity®), 4 × 9 mm (Integrity®) and 3.5 × 12 mm (Integrity®) were implanted with a significant drop in oxygen saturation (from 90% to 80%). During the procedure there was a partial migration of the stents into the pulmonary trunk, although not action was required. During the following hours, complete displacement of the stents towards the pulmonary trunk was observed, leading the patient to her previous hemodynamic state; the patient was hence returned to the catheterization laboratory (Fig. 2A). A "Diabolo stent" was successfully implanted using a coronary stent of 3 × 9 mm (Integrity®) mounted on a coronary covered stent of 3.5 × 16 mm (GRAFTMASTER®RX, Abbott, Illinois, USA). Subsequently, two additional coronary stents (Integrity® 3.5 × 12 mm) were implanted to achieve an O₂ saturation of approximately 80%. The final angiography showed a significant reduction in duct diameter, from 4.3 mm to 3.3 mm (Fig. 2B and C). Arterial O₂ saturation and pO₂ decreased from 91% and 55 mmHg to 78% and 40 mmHg respectively after the procedure. Pulmonary overflow was resolved in the following days and the patient could be discharged home.

4. Discussion

DS is a feasible and safe alternative to surgical palliation in neonates and infants with duct-dependent pulmonary circulation [5]. Although initial results were disappointing [2], advances in the technique have led to high rates of technical success with low morbidity and mortality associated with the procedure [4,7,8]. Short-term complications include stent migration, while medium-term complications include closure/narrowing of the stented duct due to progressive neointimal growth [7,8]. To a lesser extent, the presence of pulmonary overflow has complicated the short-term progress in some patients [4,9–11].

Stent diameter is chosen on the basis of the patient's size and ductal anatomy [6,12]. Nevertheless, the main factor conditioning the stent diameter is the size of the narrowest portion of the duct. Should a smaller stent than this diameter be implanted, there is high risk of embolization. Since our first patient had a straight duct, a stent 0.7 mm larger than the minimum ductal diameter was implanted in order to decrease the risk of embolization. Conversely, given the tortuous ductal morphology of our second patient, as well as the lesser concerns regarding stent migration in this patient, it was decided to implant a stent of a size similar to the minimum duct diameter.

Patients with antenatal diagnoses of duct-dependent circulation, and in whom prostaglandin infusion has been started since birth, usually feature large ducts without stenotic areas. Therefore, certain studies recommend discontinuing PGE1 infusion between 2 and 12 h before catheterization [4,8,9,11]. However, despite this measure, there is sometimes no significant reduction in duct diameter and implantation of relatively large stents may be required. In our first case, PGE1 infusion was discontinued at the beginning of the procedure, as the patient was unable to tolerate long periods of prostaglandin suspension. In the second case, PGE1 infusion was halted 6 h prior to the procedure but without a significant reduction in ductal diameter.

Most series report cases with symptoms of pulmonary overflow after DS. Although for the vast majority, this complication is managed

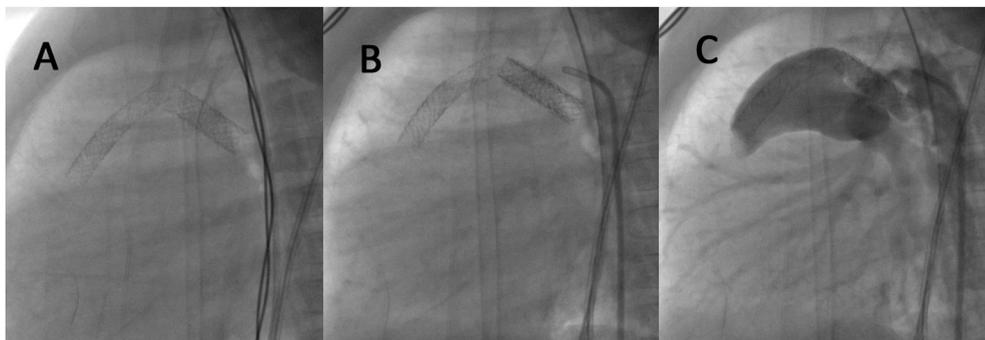


Fig. 2. A: Lateral fluoroscopic image at the beginning of third catheterization showing the ductal stent implanted in the first catheterization (Driver® Sprint 4.5 mm × 18 mm) and the displaced stents after the second procedure (one Driver® Sprint and four Integrity®). B: Lateral fluoroscopic image showing a significant reduction in ductal diameter by way of the multiple implanted ductal stents (two Driver® Sprint, six Integrity® and one GRAFTMASTER®RX). C: Final lateral aortic angiogram at the end of the third procedure.

conservatively, serious events and even death have been reported [4,9–11]. The technique of multiple stent implantation (Russian Doll technique) to treat this complication was first described by Demir et al. [13]. They presented a case of a neonate (weight 3.2 kg) diagnosed with PAIVS and signs of pulmonary overflow after arterial duct stent (4 mm) implantation. In their case, a first covered stent (3.5 mm) was implanted, however it was necessary to implant a second stent in order to further reduce the diameter of the duct, as was also the case in our two patients herein.

In our patients, since the initial stent diameter was 4.2 mm and 4.4 mm respectively, and due to the lack of covered coronary stents larger than 3.5 mm, it was decided to reduce the diameter of the duct by implanting bare metal coronary stents in order to progressively decrease the ductal diameter and avoid embolization of the covered stent. The use of the “Diabolo stent” aimed a bigger reduction in the stent’s final diameter. However, the complete dilatation of the external stent prevented this effect in both instances, thus the necessity of implanting new stents. In our second case, the bare metal stents implanted to reduce overall duct size appeared to be sufficient; however, due to stent’s displacement and the presence of residual flow across the stent’s struts, the procedure was not effective until a covered stent was ultimately implanted. In both cases, the procedure was completed when saturation dropped to 80% and the internal duct stent diameter oscillated between 3 and 3.3 mm.

5. Conclusion

The implantation of multiple coronary stents is a useful technique to reduce pulmonary blood flow in patients with severe pulmonary overflow following ductal stenting. We recommend using at least one covered stent to eliminate the residual flow across the stent’s struts. The “Diabolo stent” assembly was not shown to be useful in reducing the covered stent diameter; therefore we do not recommend such approach in this setting. Finally, the procedure should be monitored by measuring O₂ saturation, and implanting as many stents as necessary to achieve an O₂ saturation goal of 80%.

Conflict of Interest

The authors have no conflicts of interest to declare.

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