Recanalization of arterial duct is feasible, effective and its potential risks are treatable

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Background
Recanalization of arterial duct (AD) is rarely needed.

Objective
The aim of this study was to report our experience regarding the feasibility and effectiveness of arterial duct recanalization in three infants with congenital heart disease.

Methods and results
We report on three patients with decreased pulmonary blood flow after initial palliation. The first patient had pulmonary atresia (PA) and an intact ventricular septum. He needed recanalization of the AD after pulmonary valve perforation and dilatation. The second patient had PA and ventricular septal defect (VSD). His AD originated from the left subclavian artery. He required AD recanalization after palliation with a central shunt and clipping of the duct. During intervention he developed a thrombus in the stent, which was treated successfully using thrombolytic treatment. The third patient had PA and VSD. The arterial duct originated from the left subclavian artery and was recruited for spontaneous closure. Despite prostaglandin infusion, the AD remained stenotic, and recanalization and stenting were performed. All patients were successfully treated.

Conclusion
Arterial duct recanalization and stenting is a feasible and effective procedure in selected cases, and its risks are treatable. Long-term studies are required.

Keywords
Arterial duct recanalization – stent – stent thrombosis – congenital heart disease.
After femoral artery access, aortography was performed using a 4 Fr pigtail catheter in lateral and right AP view. Thereafter, the sheath was exchanged with a long one (4 Fr, 23 cm) to give better support during stent deployment. Heparin boluses were given to keep the activated clotting time (ACT) above 220 seconds. Cephazolin was also given.

**Procedure to the first case**

Aortography showed a closed AD with small indentation at the site of the ductal ampulla (figure 1a). Through a 4 Fr Ballin right 4 catheter, a floppy-II, 0.014-inch PTCA wire (Abbott, Santa Clara, California, USA) was advanced, unexpectedly easily, across the occluded AD (figure 1b). A contrast injection was done within the closed AD. The duct length was 13 mm. The catheter was withdrawn. An Integrity bare metal coronary stent (Medtronic, Minneapolis, USA), 4 mm × 15 mm was advanced over the floppy wire and deployed across the AD. The stent position was confirmed by an injection through the side arm of the sheath (figure 1c).

The saturation immediately increased from 60% to 88%. One week later, the patient was discharged on aspirin 3 mg/kg per day and clopidogrel 0.5 mg/kg per day as an antiplatelet aggregation agent. At the last follow-up, nine months later, the patient was showing normal development with a body weight of 8 kg, saturation ~93% and no complications from the antiplatelet aggregation therapy.

**Procedure to the second case**

Aortography showed a patent central shunt, but only the right pulmonary artery was visualized. A faint contrast flow to the left pulmonary artery was seen from the site of origin of the previous AD. Another hand injection was performed in the left subclavian artery, at the orifice of the clipped AD. This injection showed that there was a tiny track (14 mm in length) through the clipped duct supplying the left pulmonary artery (figure 2a). We decided to recanalize and stent the clipped AD. With the support of the 23-cm long sheath, an Integrity bare metal coronary stent (Medtronic, Minneapolis, USA), 4 mm × 15 mm, was advanced easily, positioned, and then inflated across the clipped AD. Hand injection showed that the AD was successfully recanalized and stented. The subclavian artery end of the AD remained uncovered.

A trial to deploy another stent (4 × 8 mm) to cover the remaining part of the AD led to desaturation and thrombus formation within the AD (figure 2b). The procedure was ended and the infant was transferred to the intensive care unit on heparin infusion (15 μg/kg per min). An infusion of tissue plasminogen activator (tPA) was started with close monitoring. Within two hours the saturation increased to 80%. There were no complications from the antithrombotic therapy.

**Third case**

This boy was referred to our institute at the age of 42 days with a body weight of 5.1 kg. The two-dimensional echocardiography showed pulmonary atresia with 7.0 mm right ventricle, 3.4 mm left ventricle and a patent foramen ovale. The patient was on prostaglandin infusion (10 ng/kg per min) and inotropic support. Two weeks later, aortography showed a patent central shunt with occluded AD. The left pulmonary artery branches were difficult to visualize. Two weeks later, due to persistent low saturation (~ 60%), the infant was taken to cardiac catheterization for evaluation.

**PROcedures**

All procedures were done under general anesthesia. After femoral artery access, aortography was performed using a 4 Fr pigtail catheter in lateral and right AP views. Thereafter, the sheath was exchanged with a long one (4 Fr, 23 cm) to give better support during stent deployment. Heparin boluses were given to keep the activated clotting time (ACT) above 220 seconds. Cephazolin was also given.
Arterial duct recanalization

In the third case, aortography showed pulmonary atresia, right-sided aortic arch and a barely patent AD arising from the left subclavian artery. An attempt to advance a floppy II, 0.014-inch PTCA wire (Abbott, Santa Clara, California, USA) into the AD ended in severe desaturation (50%) and bradycardia (heart rate 40 bpm). Cardiac massage and resuscitation were started. The infant recovered within two minutes. A contrast hand injection showed a completely closed AD (figure 3a). It was possible to advance the wire across the obliterated AD, and a PTCA balloon was used to reopen it. For better support, this wire was exchanged with 0.014-inch BMW wire (Abbott, Santa Clara, California, USA). As previously described, an integrity bare metal coronary stent (Medtronic, Minneapolis, USA) (4 mm × 21 mm) was deployed in the AD covering its entire length. As the AD had a curved shape, it was not easy to pull back the balloon from the stent. To overcome this situation, the long sheath was advanced and held against the subclavian end of the stent (figure 4 a, b). Thus the balloon could be withdrawn keeping the stent in place (figure 4 c). A final aortography showed a patent and well-positioned AD stent (figure 3b). The infant remained stable and was discharged within one week on aspirin (3 mg/kg per day) and clopidogrel (0.5 mg/kg per day). Ten months later, the infant had no complications from antiplatelet aggregation therapy and had normal neurological development. He is awaiting further surgical repair.

Procedure to the third case

The fluoroscopy time, contrast amount, weight, saturation and echocardiographic findings at follow-up of complications related to tPA treatment. Repeated angiography six hours later, revealed resolution of the thrombus with patency of the stent (figure 2c). The infant was discharged right after the procedure (3 mg/kg per day aspirin and 0.5 mg/kg per day clopidogrel). As the AD was patent, no further treatment was necessary. The infant had normal neurological development and was discharged within one week on aspirin (3 mg/kg per day) and clopidogrel (0.5 mg/kg per day). Ten months later, the infant had no complications from antiplatelet aggregation therapy and had normal neurological development, and underwent successful Rastelli repair.
could put less stress on the patients. Our experience with the three patients we describe shows that AD recanalization and stenting are technically feasible and effective, with midterm benefit. However, one should be aware of the potential complications of this intervention and be prepared to manage them appropriately.

In our series, two patients were not able to maintain saturation shortly after palliation (either pulmonary valve perforation and dilatation or central systemic-to-pulmonary shunt), and they required further intervention to increase pulmonary blood flow. One option was to subject them to a second surgical intervention, but the alternative was to recanalize the AD.

The benefits

In contrast to surgical shunt implantation, the ductal stent is less invasive, non-surgical and more convenient for neonates requiring multiple surgeries in the future. Additionally, after AD stenting there is better uniform growth of pulmonary artery branches, leading

all these cases are presented in table 1. Summaries of reports in the literature regarding recanalization of the AD are presented in table 2.

**DISCUSSION**

Literature review

Over the last 15 years, AD stenting has been slowly introduced as an alternative to surgical options to pulmonary shunt. Arterial duct recanalization has been described only sporadically in case reports. There was only one case series of successful AD recanalization in five out of six infants, who were older than three months of age.

Current study

We were encouraged by the few reported studies, especially when we encountered younger infants who might benefit from this intervention. Implantation of a systemic-to-pulmonary shunt was considered, but we thought that applying the concept of recanalization could provide more in the patients. Our experience with the three patients we describe showed that AD recanalization and stenting are technically feasible and effective, with midterm benefit. However, one should be aware of the potential complications of this intervention and be prepared to manage them appropriately.

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Table 1
Summary of demographic data, intervention and follow up

<table>
<thead>
<tr>
<th>No</th>
<th>Diagnosis</th>
<th>Intervention</th>
<th>Follow up</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>PA/IVS, patent AD</td>
<td>Pulmonary valvuloplasty</td>
<td>23 days 2.8 60 Integrity BMS 4/15 mm</td>
</tr>
<tr>
<td>2</td>
<td>Dextrocardia, PA/VSD, patent AD from LSA</td>
<td>Central shunt and clip of AD</td>
<td>45 days 2.9 65 Integrity BMS, 4/15 35.4 28 Stent thrombus resolved with tPA</td>
</tr>
<tr>
<td>3</td>
<td>PA/VSD, patent AD from LSA</td>
<td>None</td>
<td>50 days 3.1 55 Integrity BMS, 4/21 mm 32.2 30 Bradycardia, CPR in 2 min, respond to balloon dilation</td>
</tr>
</tbody>
</table>


Table 2
Summary of the published reports in literature about arterial duct recanalization

<table>
<thead>
<tr>
<th>STUDY</th>
<th>No. of cases</th>
<th>Age in days (weight in kg)</th>
<th>Cardiac diagnosis</th>
<th>Angiographic findings</th>
<th>Type (size) of stent</th>
<th>Final outcome of intervention</th>
</tr>
</thead>
<tbody>
<tr>
<td>Kampmann et al. 1998</td>
<td>One case</td>
<td>2 (1.89) premature TOF, disconnected LPA</td>
<td>Closed ductus arising from LSA</td>
<td>9-mm-long coronary stent dilated to 3.5 mm</td>
<td>Successful ductal recanalization and stenting.</td>
<td>Re-dilation at 235 days of age</td>
</tr>
<tr>
<td>Santoro et al. 2007</td>
<td>One case</td>
<td>3 (no wt reported)</td>
<td>Severe Ebstein anomaly</td>
<td>Completely closed and short tubular duct</td>
<td>12-mm-long coronary stent dilated to 3.5 mm</td>
<td>Successful recanalization and ductal stenting.</td>
</tr>
<tr>
<td>Kothari et al. 2011</td>
<td>Six cases</td>
<td>90-180 (no wt reported)</td>
<td>TGA/IVS, regressed LV</td>
<td>Closed ductus arteriosus</td>
<td>Coronary stent of 12-24 mm dilated to 3-3.5 mm</td>
<td>5 out of 6 had successful recanalization and stenting. Two re-interventions</td>
</tr>
<tr>
<td>El-Segaier et al. 2014</td>
<td>Three cases</td>
<td>15-45 (2.8-3.1)</td>
<td>One case PA/IVS and two cases PA/VSD</td>
<td>Closed duct (one clipped surgically, two cases arising from LSA)</td>
<td>Integrity BMS, 4/15 mm in two cases and 4/21 mm in one case</td>
<td>Successful ductal recanalization and stenting.</td>
</tr>
</tbody>
</table>

to balanced flow and development of both lung vascular networks.\textsuperscript{13,14} Potential technical difficulties

During crossing of the AD, irritation to the ductal intima may result in sudden spasm and total occlusion of the duct.\textsuperscript{3,6} This inclusion can be life-threatening, as we experienced in the third case. Another potential complication is tearing or dissection of the ductal wall by the wire or catheter. Thrombus formation within the stent is another serious risk. It may happen immediately during the procedure or later despite thrombosis prophylaxis with anti-platelet aggregation therapy. The recently occluded stent can be reopened by the use of a thrombolytic agent or by dilatation with a non-compliant balloon at higher pressure.\textsuperscript{3} Additionally, stent embolization and migration may occur during deployment.\textsuperscript{3}

The growth and morphology of pulmonary artery branches may get distorted or even totally obliterated after ductal stenting.\textsuperscript{14} Two of our cases (cases 2 and 3) had moderate stenosis of the left pulmonary artery from the start. With ductal stenting we could abolish the stenosis and save the vessels from total occlusion. Later, at the time of corrective surgery, these vessels could be reconstructed and dilated.

Midterm thrombosis prophylaxis therapy

Different authors have suggested thrombosis prophylaxis after AD stenting by a combination of aspirin and clopidogrel. We used clopidogrel at 0.5 mg/kg per day.\textsuperscript{9} A recent study challenged the need for such a “high” dose and suggested 0.2 mg/kg per day instead.\textsuperscript{15} A multi-centre, double-blind, randomized study published in 2013 disputed even the need to add clopidogrel to the usual aspirin treatment.\textsuperscript{16}

Limitations of the study

We are aware that this is a relatively new technique and the number of patients is still too small for drawing general conclusions. Therefore, larger patient series and long-term follow-up are warranted.

Conclusions

Arterial duct recanalization and stenting is feasible and effective in supplying an additional blood source to the pulmonary circulation. The procedure has some risks, but they can be minimized and treated successfully. Large patient series and long-term studies are required to evaluate the potential benefits of such intervention.

Acknowledgement

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References


