

## Case Report

## Severe Hemolysis Complicating Transcatheter Occlusion of a Patent Ductus Arteriosus: The Importance of Elimination of Residual Flow

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## Key words:

**Congenital heart disease in adults, patent ductus arteriosus, hemolysis, Amplatz Goose Neck Snare, Amplatz Duct Occluder.**

Transcatheter closure of a patent *ductus arteriosus* (PDA) is an established technique that may rarely be complicated by severe intravascular hemolysis due to residual shunt. Although conservative management has been reported, the usual approach is to invasively eliminate the residual shunt. We report a case where misjudgment of PDA size and subsequent attempted closure with only one coil led to incomplete closure and residual shunt, which was complicated by severe intramuscular hemolysis. Effectively closing the PDA with an Amplatz Duct Occluder resulted in immediate resolution of the hemolysis.

Closure of a patent *ductus arteriosus* (PDA) is indicated to prevent bacterial endarteritis, congestive heart failure, and/or pulmonary vascular disease.

Percutaneous transcatheter closure of a patent arterial duct was first performed in 1967 by Porstmann et al.<sup>1</sup> This technique has been widely used since the early 1990s.<sup>2</sup> Nowadays, transcatheter closure of a PDA with Gianturco coils and occlusive devices has become the treatment of choice at many institutions, since it is safe as well as cost-effective<sup>3,4</sup> and offers considerable advantages over surgical ligation.<sup>2,5</sup> Moreover, recent advances in device technology allow closure of virtually all PDAs, regardless of size or configuration, except in premature infants.<sup>2,6</sup>

Despite these advantages, transcatheter closure of the PDA has rare complications, one of which is severe intravascular hemolysis due to residual shunt. We report a case where misjudgment of PDA size and underestimation of residual shunt

due to a dilated left pulmonary artery resulted in severe intravascular hemolysis.

### Case presentation

A 55-year-old woman was referred to our institute for elimination of residual shunt after incomplete percutaneous transcatheter closure of a PDA with a coil, in another hospital, which was complicated by severe intravascular hemolysis.

The patient had suffered from fatigue, worsening dyspnea on exertion and palpitations for the last two years. PDA was diagnosed and the patient was admitted to the referring hospital for evaluation and treatment.

Clinical examination at that time revealed moderate jugular vein distension and a grade III/VI continuous murmur in the 2nd and 3rd interspaces to the left of the sternum. The electrocardiogram was normal and her laboratory tests were normal apart from mild hypercholesterolemia (220 mg/dl).

Her echocardiogram revealed a PDA with a moderate shunt. The main pul-

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monary artery was dilated and the estimated pulmonary pressure by Doppler was elevated (PASP ~ 50 mmHg). The right ventricle was of normal dimensions and the left ventricle was mildly enlarged.

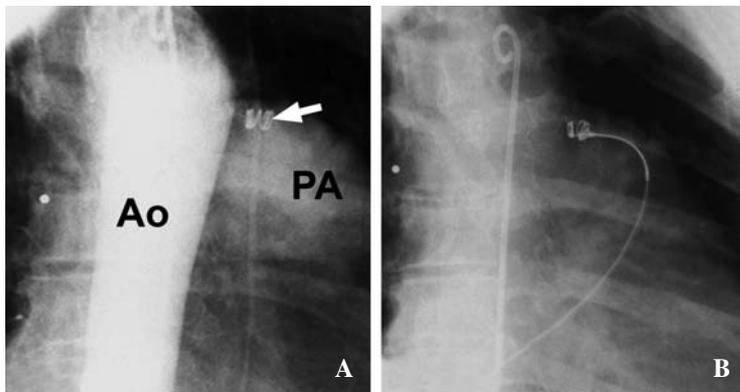
At the referring hospital, the calculated pulmonary-to-systemic blood flow ratio (Qp:Qs) was 1.7:1. At that time the PDA size was considered to be rather small and thus suitable for closure with a coil. PDA closure was attempted with a 5 mm diameter Flipper® PDA Closure Detachable Coil with 5 loops (IMWCE-5-PDA5, Cook Inc.). An aortogram, performed 10 minutes after the delivery of the coil, demonstrated what was believed to be mild residual flow due to the aneurismatic pulmonary artery.

On the following day, severe red discoloration of the patient's urine was observed. Routine urinalysis showed significant hemoglobinuria with 5-6 red blood cells /hpf. Blood tests revealed a significant drop in serum hemoglobin (14.1→11.0 g/dl), low haptoglobin (7.9 mg/dl) and elevated SGOT (149 IU/l), LDH (1781

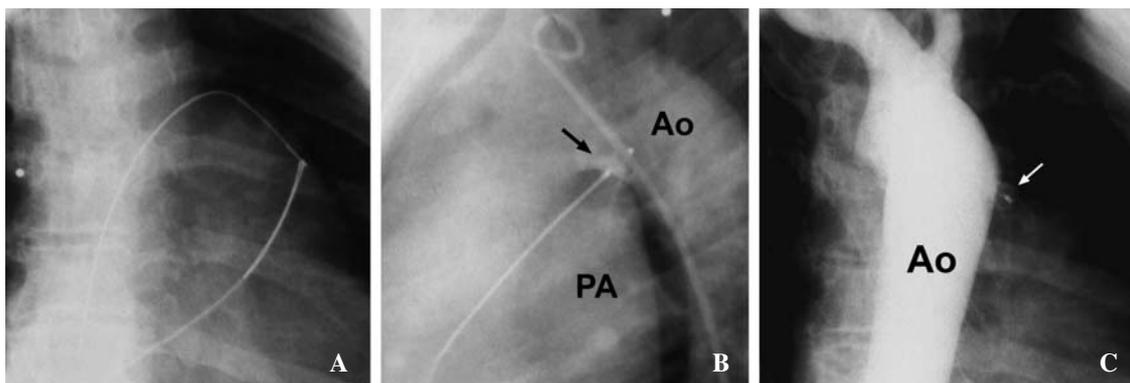
U/l), and total and indirect hemoglobin (2.48/1.7 mg/dl). The peripheral blood smear showed fragmented erythrocytes. Intravascular hemolysis was attributed to red blood cell destruction due to the residual shunt. Our institute was contacted for possible reintervention to eliminate the residual shunt.

On admission to our hospital, the patient was hemodynamically stable and was transferred directly to the catheterization laboratory.

A proximal descending thoracic aortic angiogram was performed, which showed part of the coil protruding into the left pulmonary artery and a significant residual jet through the PDA (Figure 1). Using the antegrade approach, a 25 mm Amplatzer Goose Neck Snare catheter (ev3 Inc., Plymouth, MN, USA) was advanced into the left pulmonary artery. Due to the aneurismatic pulmonary artery, difficulties arose during the attempt to entrap the coil. Finally, the snare was gently closed around the protruding edge of the coil and the coil was retrieved (Figure 2).



**Figure 1.** A. Descending aortogram in the posterior-anterior projection, demonstrating significant residual shunt following occlusion of a patent ductus arteriosus (PDA) with a PDA detachable coil. B. Cineangiogram frame in the posterior-anterior projection, demonstrating snaring of the coil via the left pulmonary artery using an Amplatzer Goose Neck Snare catheter. PA – pulmonary artery; Ao – aorta. The arrow indicates the position of the coil.



**Figure 2.** A. Cineangiographic frame in the posterior-anterior projection. An Amplatzer Goose Neck Snare catheter has captured the distal end of a straight tip 260 cm long 0.035" guidewire that was passed retrogradely through the patent ductus arteriosus into the main pulmonary artery. B. Descending aortogram in the lateral projection with the Amplatzer Duct Occluder still attached to the delivery cable, confirming a good position with no residual shunt. C. Repeat descending aortogram after the release of the device, demonstrating complete closure with good device position. PA – pulmonary artery; Ao – aorta. The arrow indicates the position of the Amplatzer Duct Occluder.

After removal of the coil, the PDA was recrossed retrogradely by a 0.035", 260 cm guidewire. The previously used snare was then advanced through the femoral vein sheath into the main pulmonary artery, the guiding wire was snared and retracted out of the femoral vein sheath creating a through-and-through wire.

Due to difficulties in measuring the diameter of the PDA by aortography, balloon sizing was performed from the venous side using a balloon-tipped end-hole catheter. The PDA diameter was estimated to be 6-7 mm. Based on this diameter we decided to use a 10/8 Amplatzer Duct Occluder (ADO). Under fluoroscopic guidance, the ADO was advanced into the descending aorta, where the retention disc was deployed. After device placement a repeat aortogram 10 minutes later revealed no residual shunts (Figure 2).

The immediate postprocedure course of the patient was uncomplicated. By the next day, the clinical and laboratory signs of hemolysis were alleviated. She remained stable and was discharged on the third post-procedure day. At one month follow up a 2D-echocardiogram with color Doppler revealed no residual flow through the PDA and no obstruction to the left pulmonary flow. The hemoglobin and indices of hemolysis were within normal limits.

## Discussion

Transcatheter patent *ductus arteriosus* closure is a safe and effective treatment. Nowadays, the most commonly used devices are the different types of coils and the Amplatzer PDA device.

In the case we presented, as a result of misjudgment of the PDA size, PDA occlusion was attempted with a Flipper® detachable embolization coil (Flipper Occluder System – IMWCE-5-PDA5).

This coil has the advantage of retrieval and repositioning in case it is not appropriately positioned on the first attempt. Apart from that, coils can be used in small PDAs and in low weight patients (infants). The easy technique of implantation, the short procedure and fluoroscopy time, the high rate of successful occlusion and the low rate of complications are some more advantages. Furthermore, only a short hospitalization is warranted and coils cost less compared to the Amplatzer device, even when multiple coils are deployed. However, the Flipper coil is less bulky and stiff than standard Gianturco coils and has fewer Dacron fibers; thus, achieving complete occlusion with a single coil is less likely.<sup>2</sup>

Major complications of transcatheter PDA closure include hemolysis after incomplete closure, device embolization to a systemic or pulmonary artery, infection, left pulmonary artery obstruction, thoracic aorta obstruction and femoral arteriovenous fistula formation.<sup>5-7</sup>

Hemolysis is thought to result from red blood cell mechanical injury when a high-velocity residual jet comes in contact with the metallic surface of the occluding coil or device. It is more common after coil closure than with other devices and some authors recommend not leaving any residual shunts even if multiple coils are required.<sup>5-8</sup> Significant hemolysis, however, is a rare complication and its risk is greatest in patients of younger age, lower body weight and with relatively large ducts.<sup>8</sup> Hemolysis usually develops in the first 24 hours postprocedure, but may occur weeks later.<sup>8</sup> When severe it may result in anemia, jaundice, renal failure and coagulopathy.<sup>5-8</sup>

The usual approach to the patient with significant intravascular hemolysis is stabilization, protection against renal damage, and eradication of residual flows. This is accomplished by correcting anemia through blood transfusions, iron and folate supplementation, avoiding dehydration and acidosis by precise fluid management, and by inducing alkaline diuresis.

Elimination of residual flows may be achieved by using additional coils, adding coils to a device, replacing coils with an occluding device, or as second choice by surgical ligation, with or without removing the previously implanted device.<sup>3,5,6,8</sup>

In our case, we decided to replace the coil with an Amplatzer Duct Occluder. For coil removal we used an Amplatz Goose Neck Snare, which was also used to facilitate antegrade closure of the PDA with the Amplatzer device. The Amplatz Goose Neck Snare has its loop at a right angle to the catheter, which made the capturing of the coil easier. It is used to place, replace, reposition, or to remove devices during interventional procedures.<sup>9</sup>

We used the Amplatzer Duct Occluder (AGA Medical Corporation, Golden Valley, MN, USA) to occlude the PDA. It is recommended for nonsurgical duct closure independent of the shape and size of the duct. The ADO stenting results in virtually a 100 percent occlusion rate, obviating the need for multiple device implantations and reducing the risk of embolization. Another advantage is that it can be retracted and redeployed several times. However, rare complications have been reported and are the same as with coils, i.e. device embolization, left pulmonary

stenosis, protrusion into the aorta, and haemolysis.<sup>2,10</sup>

Apart from the invasive approach, in patients with mild hemolysis conservative medical treatment has been reported with administration of haptoglobin to protect against renal damage and antiplasmin infusion to promote thrombus formation, since many residual shunts resolve spontaneously.<sup>5</sup> However, the consensus is to manage this complication invasively.

In conclusion, this case highlights the importance of the “no residual shunt” strategy<sup>6</sup> to minimize the occurrence of severe hemolysis. Furthermore, it points out the significance of performing a thorough analysis of the morphology and size of the PDA, in order to select the appropriate type of closure device (one device or coil, multiple coils or a combination of a device and coils). In other words, the knowledge, skill and experience of the catheterization team will ensure less procedure failures, less residual shunts and less obstructive or other complications.

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