Hemolysis associated with coil occlusion of the arterial duct

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Abstract  Severe mechanical hemolysis occurred in a 22 month old girl after placement of a 38-5-10 coil in the arterial duct. She had previously undergone percutaneous closure using the Rashkind technique 14 months before insertion of the coil, but remained with a moderate residual shunt. Surgical removal of the devices and division of the duct were required to abolish the hemolysis.

Key words: Patent ductus arteriosus; coil occlusion; hemolysis

Transcatheter closure of the arterial duct using the Rashkind technique has become a well established alternative to surgical treatment. Residual shunts are not uncommon after the procedure, although persistent shunting can be managed by placement of a second umbrella. Most recently, successful occlusion of residual shunts has also been achieved using coil embolization. Hemolysis occurs only rarely after umbrella closure of the arterial duct. Reported management for this complication includes surgical or transcatheter removal of the device, ligation of the duct over the device, or implantation of a second umbrella. Hemolysis following placement of coils is probably even less common. We describe here a case of severe mechanical hemolysis resulting from implantation of a coil in the duct in order to abolish late residual shunting following initial closure with an umbrella.

Case report
A symptomatic 12-month-old girl (8 kg) with clinical and echocardiographic findings consistent with a hemodynamically significant persistent arterial duct was admitted for transcatheter occlusion using the Rashkind technique. The lateral aortogram revealed a tubular duct with a minimum internal diameter of 3.8 mm. A 12 mm umbrella was placed in a good position without difficulty. Another aortogram performed 20 minutes after the implantation showed moderate residual shunting (Fig. 1). Doppler echocardiography 24 hours later confirmed this finding.

Fourteen months later, she was asymptomatic but a continuous murmur was still heard below the left clavicle. Serial doppler echocardiography performed 3, 6 and 14 months after the procedure showed persistence of the initial moderate residual leak. In view of residual shunting, another cardiac catheterization was undertaken. An aortogram confirmed the moderate residual shunt. A 38-5-10 coil (Cook) was easily implanted over the umbrella using a retrograde approach. The aortic end of the coil, however, remained protruded minimally towards the aorta (Fig. 2). A repeat aortogram demonstrated little change in the residual shunt, and this finding was confirmed by doppler echocardiography performed 24 hours later.

After 4 weeks, the child re-presented with pallor and a history of hematuria and jaundice. The hemoglobin was 67 g/l, reticulocytes count was 15%, and red cell fragments were noted on blood film. Microscopic examination of the urine showed hemoglobin but no red blood cells. The child was readmitted and was initially managed conservatively. After transfusion of blood, the
Figure 1.
Lateral aortogram
A) Tubular duct with a minimum internal diameter of 3.8mm
B) Moderate residual shunt after implantation of umbrella 12

Figure 2.
Lateral aortogram, coil 38-3-10 implanted over the umbrella
C) View of the coil implanted.
D) Maintenance of moderate shunting. Aortic end of the coil protruded towards the descending aorta.
hemoglobin concentration raised to 115 g/l. A repeat echocardiogram indicated that the aortic end of the coil was situated further into the aorta than anticipated, and doppler study continued to show a significant leak through the duct. After 72 hours, the hemoglobinuria was no better and the hemoglobin had fallen again to 93 g/l. In view of the continued hemolysis, another cardiac catheterization was performed. The aortogram confirmed the sub-optimal position of the coil and the residual leak. An attempt to retrieve the coil using the venous approach was made but it was impossible to remove the coil without dislodging the umbrella. A second coil (38-5-8; Cook) was therefore implanted using a retrograde approach. Unfortunately, this second coil migrated to the pulmonary arteries and remained connected to the first coil, swinging inside the left pulmonary artery. Because of the imminent risk of embolization, it was immediately and easily retrieved. The child was then referred for surgical treatment.

The duct was exposed through a left lateral thoracotomy and the devices were easily palpable within it. The left pulmonary artery and the descending aorta were side clamped at the level of the duct. The duct was partially incised and the coil was removed. The duct was then divided leaving the umbrella (which was endothelialised and firmly attached to the wall of the duct) towards the pulmonary side. The free aortic side was sutured and a further suture was placed in the pulmonary end. The portion of the duct with the umbrella was totally excised and the suture line on the pulmonary side was completed. There was no evidence of infection. Postoperative recovery was uneventful with no further hemolysis. The child was discharged home 5 days later with a hemoglobin concentration of 145 g/l.

**Discussion**

Hemolysis is a rare complication following umbrella closure of the arterial duct. It is estimated to occur with an incidence of 0.5% and generally manifests soon after the procedure. It is always associated with residual shunts and occurs irrespective of whether the device is well positioned. Hemolysis after implantation of coils in the duct is probably even less common. In our patient, as was speculated by others, the residual shunt following closure using the umbrella may have been exacerbated by underestimating the size of the duct, leading to insertion of an inappropriate device (too small -12 mm). Residual shunts, of course, can be managed by implantation of a second umbrella. Most recently, however, there has been a movement to use coils for occlusion of the patent duct, including also those with residual leaks after surgical ligation or occlusion with umbrellas. Based on these, and on our own experiences, we decided to use a coil to abolish the residual flow in our patient. The procedure itself was simple, but the aortic end of the device remained slightly protruded towards the descending aorta. We postulate that this fact was responsible for the development of the hemolysis: The distal end of the coil faced directly the high pressure jet of blood from the aorta, causing damage to the red blood cells. It is unlikely that the residual leak in itself was responsible for the hemolysis, since it had been present prior to insertion of the coils, and its magnitude was not much affected by coil implantation. Probably, we selected a coil of inadequate size to occlude the residual leak and placed it in a wrong position. With the coil of 10 mm helical diameter, the 5 cm length available produced just one and a half loops. As one loop was accommodated inside the duct, a considerable length of unlooped coil remained positioned inside the descending aorta, leading to the disturbance in flow. A coil with a smaller helical diameter (5 or 8 mm), but with a larger length (sufficient to produce at least three to four loops) would have been a better choice. Alternatively, it might have been preferable to use a detachable coil, which have been much easier to site correctly.

Since there is no unanimity as yet to what exactly causes hemolysis, several approaches have been proposed to manage the problem. Mild hemolysis may require no intervention, and even severe hemolysis has subsided with conservative therapy. Catheter or surgical removal of the device is an obvious approach. Some have advocated implantation of a second device as the treatment of choice to stop hemolysis.

These authors believe that abolition or marked reduction of residual flow through the duct is more important rather than removal of the device itself. One case of hemolysis associated with persistent flow after coil occlusion was managed by retrieval of the coil and implantation of a Rashkind umbrella. Taking all these observations into consideration, we offered to our patient a initial short period of conservative treatment, but the hemolysis failed to resolve. Retrieval and implantation of a second device were both attempted, but without success. Surgery was then the only way to manage the problem. Surgical removal of the devices, with division of the duct, was feasible...
although it was technically difficult, requiring side clamping of the aorta and left pulmonary artery. In some cases, a short period of cardiopulmonary bypass may be inevitable.

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References